

PHYSICAL ACTIVITY AND FITNESS IN CHILDREN WITH DEVELOPMENTAL COORDINATION DISORDER

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ABSTRACT

Introduction: Developmental coordination disorder (DCD) is a prevalent condition characterized by poor motor proficiency that interferes with a child's activities of daily living. Children with DCD often experience compromised health-related fitness components such as cardiorespiratory fitness (CRF).

Purpose: To better understand the physical activity and fitness characteristics of children with probable DCD (pDCD), with a particular focus on CRF. Specifically: (1) to present a synopsis of current literature; (2) to determine the longitudinal trajectories of CRF; (3) to compare the submaximal CRF of children with and without pDCD.

Methods: A comprehensive, systematic literature review was conducted of the recent available data on fitness and physical activity and pDCD (Chapter 2). This review provided the background for the other two studies included in this thesis. In Chapter 3, a prospective cohort design was used to assess how CRF in children with pDCD changes over time (56 months) relative to a group of typically developing controls. Using a nested-case control design, 63 subjects with pDCD and 63 matched controls from the larger sample were recruited to participate in the lab-based component of the study (Chapter 4). In this investigation CRF was examined using the oxygen cost of work (VO_2) during an incremental test on a cycle ergometer.

Results: The literature review showed that fitness parameters, including CRF and physical activity levels, were consistently reduced in children with pDCD. Chapter 3 demonstrated that the difference in CRF between children with pDCD and typically developing children is substantial, and that it tends to increase over time. Results from

VO₂ assessments showed that children with pDCD utilized more oxygen to sustain the same submaximal workloads compared to typically developing children.

Conclusions: Findings from this thesis have made several important contributions to our understanding of children with pDCD. Since differences in CRF between children with and without pDCD tend to worsen over time, this adds to the argument that interventions intended to improve CRF may be appropriate for children with motor difficulties. This thesis also presented the first evidence suggesting that DCD involves higher energy expenditure, and could help explain why children with pDCD perform poorly on tasks requiring CRF.

Keywords: Developmental coordination disorder, cardiorespiratory fitness, physical activity, prospective cohort, oxygen cost.

PREFACE

This thesis was prepared in an integrated article format. Sections of this thesis have been or will be published as multi-authored manuscripts in peer reviewed journals. Specifically, this manuscript is comprised of three papers investigating the broad topic of physical activity and fitness in children with DCD. The first article is a systematic review of the literature on physical activity and fitness patterns in children with DCD (Chapter 2). The second article examines longitudinal trajectories of cardiorespiratory fitness of children with and without DCD in a prospective cohort design (Chapter 3). The third article reports on results of a laboratory based nested case-control study of cardiorespiratory fitness in a sample of children with DCD and matched, typically developing controls (Chapter 4).

Published Manuscripts:

Chapter 2: Rivlis I, Hay J, Cairney J, Klentrou P, Liu J, Faught BE. (2011). Physical activity and fitness in children with developmental coordination disorder: A systematic review. *Research in Developmental Disabilities*. 32: 894–910.

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LIST OF ABBREVIATIONS

α	Alpha
BMI	Body Mass Index
BOTMP-SF	Bruininks Oseretsky Test of Motor Proficiency
CRF	Cardiorespiratory Fitness
CSAPPA	Children's Self-perception of Adequacy in and Predilection for Physical Activity
DCD	Developmental Coordination Disorder
pDCD	Probable Developmental Coordination Disorder
FFM	Fat Free Mass
HR	Heart Rate
MABC-2	Movement Assessment Battery for Children, Version 2
PA	Physical Activity
pDCD	Probable DCD
PHAST	Physical Health Activity Study Team
PE	Physical Education
PQ	Participation Questionnaire
RER	Respiratory Exchange Ratio
RPE	Ratings of Perceived Exertion
VO ₂ peak	Peak volume of oxygen
FFM	Fat Free Mass

CHAPTER 1 – Introduction

1.1. Introduction to the disorder

Developmental coordination disorder (DCD) is a neurodevelopmental condition affecting approximately 5-8% of school aged children (APA 2000, Gubbay, 1975; Henderson & Hall, 1982; Gillberg & Kadesjo 2003; Cermak & Larkin, 2001). The most prominent feature of DCD is a marked impairment in the development of motor coordination that can affect the performance of daily activities such as writing, handling small objects, and engaging in physical activity like riding a bike or catching a ball (Polotajko et al. 2005). The movement difficulties experienced by children with DCD are not due to a pervasive developmental disorder or other intellectual or neurological impairments that could explain the deficits. It is generally believed that DCD is a chronic impairment that persists into adulthood (Barnhart 2003; Cantell et al. 1994). Increasingly, literature focusing on the motor deficiencies experienced by children with DCD has revealed the heterogeneity of this condition, with some children having challenges with fine motor skills, gross motor skills, or both, and with some children having more profound and complex difficulties than others (Green et al. 2008, Hoare, 1994). While our understanding of this condition has markedly improved over the last few decades; there are still many areas that require further exploration.

1.2. Diagnosis and Assessment

The term Developmental Coordination Disorder is reasonably current. However, the condition has been recognized in some form in the literature over the past several decades, often describing children as “awkward”, “clumsy”, or having “movement

difficulties” (Geuze et al. 2001). As early as 1937, children with mild motor problems have garnered interest in pediatric medical research (Orton, 1937). Orton used the term “developmental” to emphasize the challenge children with this disorder face in developing motor skills or reaching age appropriate milestones. Others have referred to DCD as a “deficit in the acquisition of skills” that require coordinated movement (Hall, 1988 p.375). Additional terms such as “developmental dyspraxia”, “apraxia”, and “minimal brain dysfunction” have also frequently appeared in the literature. However, since a more refined definition was released in 1987, the term developmental coordination disorder has gained popularity in recent literature (APA, 1994; Geuze et al. 2001). Following an international consensus meeting held in London, Ontario in 1994, the research community has agreed upon the term developmental coordination disorder primarily to standardize research efforts in the field, and in practice to help identify children with motor challenges (Missiuna & Polatajko, 1995). It has also been generally accepted that the diagnostic criteria outlined in the Diagnostic Statistical Manual (DSM-IV, 1994; pp 54-55) should be used in diagnosing developmental coordination disorder. These criteria include: (DSM-IV, 1994):

- A. Performance in daily activities that require motor coordination is substantially below that expected given the person’s chronological age and measured intelligence. This may be manifested by marked delays in achieving motor milestones (e.g., walking, crawling, sitting), dropping things, “clumsiness,” poor performance in sports, or poor handwriting.
- B. The disturbance in criterion A significantly interferes with academic achievement or activities of daily living.

C. The disturbance is not because of a general medical condition (e.g., cerebral palsy, hemiplegia, or muscular dystrophy) and does not meet criteria for a pervasive developmental disorder.

D. If mental retardation is present, the motor difficulties are in excess of those usually associated with it.

The International Classification of Disease (ICD-10), although less frequently used in the literature, is another diagnostic system that uses the term “specific developmental disorder of motor function” to refer to DCD (WHO, 1996). While the DSM-IV is a criteria-based diagnostic approach, the ICD-10 recommends norm-referenced standardized testing to diagnose the disorder. According to the diagnostic guidelines of ICD-10, assessment of children should involve individually administered standardized tests of fine and gross motor movement (WHO, 1996). Since 1994, the term developmental coordination disorder has prevailed in the literature. However, the description of the diagnostic criteria provides opportunity for varying interpretations, and adherence to the selection criteria has not been consistent (Geuze et al. 2001).

Identifying children with DCD in clinical practice and in the research setting can sometimes be challenging, not only due to the heterogeneity of the disorder, but also because of the various measurement tools used to assess DCD status. The Bruininks Oseretsky Test of Motor Proficiency (BOTMP) is a screening tool for DCD, where test items are organized into eight categories (in the original version), representing a standardized, norm-referenced measure that can be used by therapists and researchers in clinical and school settings (Bruininks, 1978; Bruininks & Bruininks, 2005). Another widely used measure is the Movement Assessment Battery for Children (MABC), which

produces both normative and qualitative measures of movement competence, manual dexterity, ball skills, and static and dynamic balance (Henderson & Sugden, 1992). A Canadian instrument, the Developmental Coordination Disorder Questionnaire (DCDQ), has also been applied in some studies, albeit less frequently (Wilson, 2005; Cantell et al. 2008). Cairney et al. (2009) showed that the short form of the BOTMP is a reasonable alternative to case identification when clinical assessment with the MABC is not feasible, with a positive predictive value of 0.88. While in other studies, the BOTMP and the MABC have shown moderate to high agreement (67-82%) in distinguishing those with and without DCD, highlighting the potential misclassification of the available test instruments is noteworthy (Portney & Watkins 2000; Crawford et al. 2001).

1.3. Deficits and Prognosis of Developmental Coordination Disorder

DSM-IV emphasizes that DCD involves a marked impairment in the development of motor coordination, which must interfere with academic achievement or with activities of daily living. A general medical condition that might explain the trouble with movement control must be excluded (e.g., muscular dystrophy or cerebral palsy). The difficulties experienced by children with DCD have been well documented (Polatajko 2005; Dunford et al. 2005). Analyzing the literature on DCD, Macnab et al. (2001) found five different subtypes of DCD, pointing to the heterogeneity of the condition. Each subtype is characterized by deficits in fine motor, gross motor, kinesthetic, visual, or dexterity skills. Examples of deficits include children who have difficulty with running, holding a knife and/or fork, buttoning clothes, or playing ball games (Wilms Floet & Maldonado-Duran, 2010). DCD may manifest as challenges with gross motor movement

such as poor balance, clumsiness, dropping or bumping into things, catching, kicking, running, jumping, hopping, and/or persistent difficulty with fine motor control (e.g., writing, cutting, printing) (Blank et al. 2012). The acquisition of motor skills may also be affected. While disturbance in criterion I of the *DSM-IV*: significantly interferes with activities of daily living, may be evident in difficulties with self-care (e.g., dressing), academic performance, leisure and play activities (Blank et al. 2012).

Many children with motor coordination difficulties also have coexisting conditions. Some of the most common ones are learning difficulties as well as attention deficit hyperactivity disorder (Baerg et al. 2011; Blank et al. 2012). Children with DCD often report negative feelings about themselves, low perceived competence in the physical domain, and reduced motivation to participate in physical activities (Hay & Missiuna, 1998; Losse et al., 1991; Silman et al., 2011). In the past, it was believed that children with DCD would outgrow their motor difficulties (Sellers, 1995). However, longitudinal studies have shown that the motor challenges of children with DCD usually persist into adolescence and adulthood (Cantell et al., 1994; Losse et al., 1991).

1.4. Fitness, Physical Activity and Developmental Coordination Disorder

In recent years, a growing issue of interest has been the physical health of children with DCD. In light of the increasing prevalence of hypoactivity and cardiovascular disease risk factors observed in children and adolescents, those with compromised motor proficiency may experience additional challenges engaging in physical activity. One of the many consequences of reduced physical activity is that health-related fitness components such as cardiorespiratory fitness (CRF) are

compromised (Cairney et al., 2007; Hands, 2008). In fact, research exploring the fitness and physical activity patterns of children with poor motor proficiency has provided a rather alarming risk profile for cardiovascular disease, due to higher percentage of body fat, decreased aerobic capacity, and generally decreased participation in physical activity (Cairney et al., 2007; Faught et al., 2005; Schott et al., 2007). Children with DCD may avoid physical activity because they often lack a sense of competence when participating in activities compared to typically developing children (Poulsen, 2007a). The consequences of this avoidance may include not only decreased opportunity to develop overall physical fitness, but also social and emotional challenges such as depression and social isolation (Bouffard et al., 1996; Bar-Or & Rowland, 2004)

Many gaps in the literature still exist. In particular, large scale, longitudinal, studies that quantify risk are still lacking. While the body of knowledge examining various aspects of physical activity, fitness, and health of children with DCD has been steadily increasing, no systematic review of the recent evidence has been published. This thesis aims to address the need for a recent examination of physical activity and fitness in children with DCD. Therefore, a comprehensive systematic review of the literature will be valuable in synthesizing the recent available data on fitness and physical activity in children with DCD, in understanding the extent of the differences between typically developing children and those with the disorder, and to inform future research efforts and current clinical practice. Previous research has demonstrated that higher levels of aerobic fitness are associated with a healthier cardiorespiratory profile in children and adolescents (Ortega, 2008; Twisk, 2002). Conversely, poor CRF early in life may result in the development of cardiovascular diseases in later life (Berenson, 2002). Considering

the sedentary lifestyle reported in children with DCD, and the important influence of CRF on overall health, this thesis will consider the influence of DCD on the longitudinal trajectory of CRF.

In order to gain a better understanding of the factors associated with poor CRF performance in children with DCD, this research will extend previous work by Silman et al. (2011) that purports that DCD may involve higher energy expenditure. Silman and colleagues suggested that differences in peak oxygen uptake (VO_{2peak}) could be accounted for by the negative consequences of DCD, such as poor movement patterns resulting in higher energy expenditure and higher levels of fatigue. Although they were unable to test the submaximal aerobic differences, the authors speculated that even slightly compromised movement efficiency in children with DCD may have contributed to increased energy demands at various levels of physical workload. We sought to extend this speculation in the current study. Understanding why children with DCD perform more poorly on tests of CRF can provide insight for future research and the design of appropriate interventions.

1.5. Physical Health Activity Study Team

Data collected through the Physical Health Activity Study Team (PHAST) study will be utilized to address the identified research gaps. The PHAST study incorporated a prospective surveillance of children registered in grade four in 2004 from the District School Board of Niagara to examine their fitness and physical activity patterns, motor coordination deficits, and corresponding risks for cardiovascular disease. The research presented in this thesis is the culmination of this six year longitudinal examination. A

total of 2278 children enrolled in Grade 4 at baseline (representing 75 of 92 possible schools) agreed to participate in the PHAST annual school-based health assessments. From within this larger cohort, a nested case-control examination of 63 subjects with probable DCD and 63 age, gender and school location matched controls participated in a lab-based investigation. Recruitment of subjects, procedures and data collection methods are described relative to the specific studies below.

1.6. Objectives of the Thesis

The overall purpose of this thesis was to better understand the physical activity and fitness characteristics of children with DCD. Given the gaps in the literature outlined in the previous section, and the available PHAST dataset, this research aims to address some of these identified gaps. The body of this thesis is comprised of three manuscripts that have been published or submitted for publication in peer-reviewed journals. The manuscripts are reproduced in Chapters 2 to 4. Outlined below are the specific objectives for each study:

- 1) Systematically review the existing literature on children with DCD in order to better understand the physical activity patterns and fitness characteristics of this population, and address areas requiring further research.
- 2) Determine the longitudinal trajectory of cardiorespiratory fitness in children with DCD and delineate factors that influence this relationship.
- 3) Compare the submaximal aerobic performance of children with and without DCD on a $\text{VO}_{2\text{max}}$ test, in order to examine the differences in oxygen cost at submaximal workloads.

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CHAPTER 2 - Physical Activity and Fitness in Children with Developmental Coordination Disorder: A Systematic Review¹

2.1. Introduction

Developmental coordination disorder (DCD) is a neurodevelopmental condition thought to affect approximately 5-8% of school aged children (APA 2000; Gubbay, 1975; Henderson & Hall, 1982; Gillberg & Kadesjo, 2003; Cermak & Larkin, 2002). DCD is a complex disorder characterized primarily by poor motor skills that interfere with a child's activities of daily living (Cermak & Larkin, 2002). The movement difficulties experienced by children with DCD do not result from a pervasive developmental disorder or other intellectual or neurological impairments. It is not known precisely what causes DCD, although it is believed that DCD may have a genetic component (Lichtenstein et al., 2010), and/or is associated with perinatal oxygen perfusion problems (Pearsall-Jones et al., 2009), and is generally a chronic impairment that persists into adulthood (Barnhart et al., 2003; Cantell et al., 1994).

In recent years, a growing issue of interest has been the physical health of children with DCD. Considering the increasingly low levels of fitness and physical activity that are typically observed in children in the general population; children with DCD are potentially at a greater disadvantage given the nature of their disorder. Numerous studies have shown that children with DCD have on average lower fitness levels compared to their peers (Table 2-1). Daily activities that most young children engage in such as running, walking, and jumping are important for the proper development of fitness and overall health (Cermak & Larkin, 2002). However, children with DCD usually find these

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activities difficult and may experience lower desire to participate in activity, which leads to lower likelihood of participating in opportunities to develop proficient motor skills and adequate fitness levels.

Research exploring the fitness and physical activity patterns of children with poor motor proficiency has provided a rather alarming risk profile for cardiovascular disease, due to higher percentage of body fat, decreased aerobic capacity, and generally decreased participation in physical activity (Cairney et al., 2007; Faught et al., 2005; Schott et al., 2007). However, many gaps in the literature exist. In particular, large scale, longitudinal, studies that quantify disease risk are still lacking. In 2002, Hands and Larkin presented a comprehensive review of studies on physical activity and fitness in children with DCD (in Cermak & Larkin, 2002). Since then, the body of knowledge examining various aspects of physical activity, fitness, and health of children with DCD has been steadily increasing. Fitness components including body composition, cardiorespiratory fitness, muscle strength and endurance, anaerobic capacity, power, and flexibility are important in the proper development of children's health and well being. To date, no systematic review of the recent evidence regarding fitness and physical activity in children with DCD has been published. A systematic review of the literature will be valuable in synthesizing the recent available data on fitness and physical activity in children with DCD, and in understanding the extent of the differences between children with DCD and typically developing peers.

2.2. Methods

2.2.1. Selection of Studies

A systematic review of the literature was conducted to identify relevant studies reporting on physical activity and/or fitness in children with motor coordination difficulties. A search strategy was devised that combined three groups of terms, including: i) motor proficiency, ii) fitness and physical activity, and iii) age group of interest. A study's title and abstract were required to contain at least one term from each group to be considered for inclusion in the review. The first group of terms included terminology that captured motor coordination difficulties, including variations of the following: developmental coordination disorder, motor skills disorder, coordination disorder, in-coordination, clumsy, motor proficiency, motor competence, motor difficulties, and motor impairment. We employed a liberal approach in the search strategy as terms are often used interchangeably depending on study's origin, and, since DCD is a more contemporary diagnosis, in order to capture older studies. The second group of terms aimed to capture the outcomes of interest (i.e., fitness, physical activity), including variations of these terms: exercise, sport, sedentary, inactive, aerobic, anaerobic, endurance, strength, flexibility, agility, power, body composition, overweight, BMI (body mass index), adiposity, body fat. The last group of terms focused on the population of interest (e.g., children, teens, adolescents, youths, students, boys, or girls).

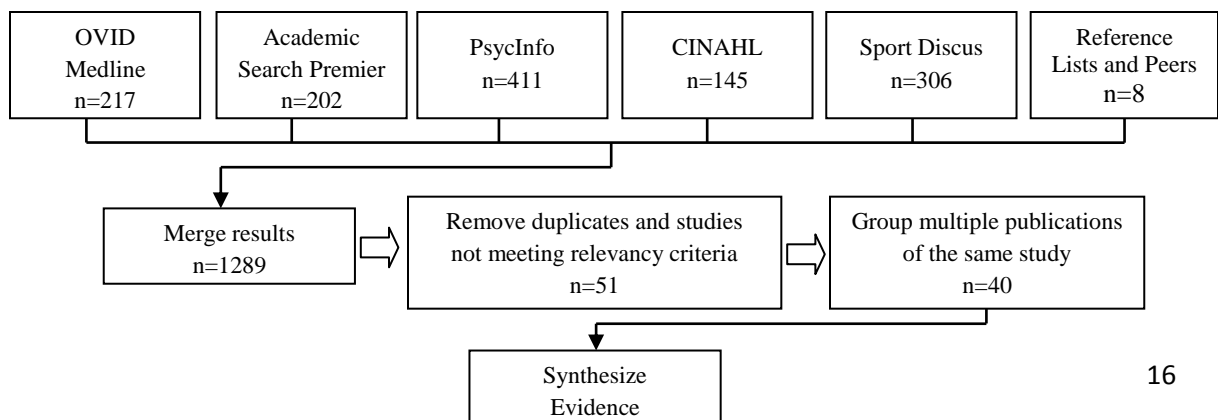
The following five electronic databases were systematically searched: OVID Medline, Academic Search Premier, Cumulative Index to Nursing and Allied Health Literature, Sport Discus, and PsycInfo. The terms used were customized for each database, so that the databases' controlled vocabulary was used whenever possible. The

search was not limited by studies' publication date. In addition, we consulted with peers in the field and manually reviewed the reference lists of pertinent papers to identify any papers not captured in the electronic database search. Only those studies found in peer reviewed journals, and those published in English were included. Our search strategy was designed to be inclusive. We sought to include all studies that would provide information about the fitness characteristics and physical activity patterns of children with motor difficulties (e.g., observational, clinical and intervention studies were included). Therefore, we did not specifically exclude studies whose objectives were to test intervention effectiveness, or to assess changes in motor proficiency over time, even if the main outcomes were not relevant to our review. Publications that did not include data on the measures of interest, namely physical activity or fitness were excluded.

2.3. Results

The search yielded 1289 potentially relevant publications (Figure 2-1). After reviewing the titles and abstracts and removing duplicates; 51 articles were identified that met our relevancy criteria. To avoid repetition, we grouped those studies that were published by the same authors in multiple papers, which narrowed the results down to a total of 40 studies that proceeded to the evidence synthesis stage.

Figure 2-1. Systematic review process



2.3.1. Study Characteristics

Studies that contributed to evidence synthesis were characterized according to study design, sample cohort, motor proficiency assessment tools, measures, fitness outcomes, and physical activity outcomes (see Table 2-1). Of the 40 studies included in the review, eight utilized longitudinal study designs, 31 articles were cross-sectional studies, and one a case-study. The follow-up durations of longitudinal studies ranged from 10 weeks to 10 years. Most studies utilized comparison groups, such as children who had definite motor difficulties versus those who were typically developing, or a mixed sample of children with varying motor skills. Most publications were descriptive or observational, while the remainder were intervention studies (n=2). Of the 40 reviewed studies, only three did not utilize comparison groups. Of these, one was a qualitative study (Mandich et al., 2003), one was a case study (Kaufman & Schilling, 2007), and the third was a longitudinal intervention study (Peters & Wright, 1999). We did not restrict inclusion based on study design or sample size, as we thought all study types can provide information. Therefore, sample sizes varied from one child in the case study published by Kaufman and Schilling (2007) to a sample of 2278 children in the articles by Cairney and colleagues (2010a,b).

Different methods of DCD case ascertainment were used in the reviewed studies. The most popular instrument, utilized in 17 studies, was the Movement Assessment Battery for Children (MABC) (Henderson & Sugden, 1992). This was not surprising, given it is the most widely used standardized motor test to screen for motor impairment in research (Wilson, 2005), and because there is evidence of reliability (test-retest) and some validation work that has been done on the measure, at least in relation to criterion

and concurrent validity (Crawford et al., 2001, Tan et al., 2001). The Bruininks Oseretsky Test of Motor Proficiency (BOTMP) (Bruininks, 1978) is one of the most popular measures used by North American researchers and health professionals to assess motor skills (Crawford et al., 2001). The BOTMP, either the long or short form of this measure, is designed to assess both gross and fine motor skills in children, and was utilized by nine of the reviewed studies. Other instruments included the McCarron Assessment of Neuromuscular Development (MAND) (McCarron, 1997), which was used in five studies. Finally, the Test of Motor Impairment (TOMI) was used in two studies. Five studies assessed motor competence using other tools specific to their study. For example, Castelli and Valley (2007) chose the South Carolina Physical Education Assessment Program (SCPEAP) motor skill testing protocols and scoring criteria in their study that involved striking a ball with paddles, basketball handling, and ball throwing tasks. The SCPEAP assessments were selected because it suited the age group of the study cohort (7-12 year olds) and the authors were able to demonstrate high inter-rater reliability in pilot testing (Castelli & Valley, 2007).

In terms of the outcomes that were encountered in the various studies, 27 included at least one fitness outcome of interest: body composition, aerobic fitness, anaerobic fitness, muscle strength, power, or flexibility. Physical activity outcomes were investigated in 22 studies, while 11 studies examined both fitness and physical activity outcomes simultaneously.

Table 2-1. Summary of studies on physical activity and fitness

	First Author (year)	Study Design	Population	DCD Assess- ment Tool	Measure(s)	Fitness Outcomes*	Physical Activity Outcomes
1	Barnett (2009)	Longitudinal (6-7 yrs follow up)	276 children with and without object control proficiency problems, mean age at follow up 16 yrs	Assessment of various motor control skills	Physical activity recall	NA	Object control proficient children became adolescents with a 10% to 20% higher chance of vigorous activity participation.
2	Bouffard (1996)	Cross-sectional	52 children with and without movement difficulties, ages 6-9 yrs	TOMI	Physical activity participation during recess	NA	DCD group was vigorously active less often, played less often with playground equipment, and generally participated less in physical activity
3	Burns (2009)	Cross-sectional	109 children with and without coordination problems; half with extremely low birth weight (ELBW), ages 11-13 yrs	MABC	CRF Muscle strength	70% of ELBW group had definite DCD. 45% of ELBW group were below the 10th percentile for VO2 peak, and had poorer strength. Poor MABC score predicted lower VO2 peak in both ELBW and comparison groups	NA
4	Cairney** (2005-2007) & Faught (2005)	Cross-sectional	590 children with and without DCD, ages 9-14 yrs	BOTMP	Body composition CRF Physical activity recall	DCD was associated with overweight and obesity, and differences persisted over time. DCD group had lower aerobic fitness scores on the Léger 20m run	DCD group participated less in organized and free play, also reported lower average enjoyment of physical education, and lower perceived adequacy for physical activity

5	Cairney (2010a,2010b)	Longitudinal (2.5 yr follow up)	2278 children with and without DCD, ages 9-10 yrs at baseline	BOTMP	Body composition CRF	Children with DCD had higher BMI and waist circumference at baseline, and these differences persisted or increased slightly over time. DCD group not only had lower VO2 peak at baseline, it declined at a much steeper rate	NA
6	Cantell (1994)	Longitudinal (10 yr follow up)	115 children with and without motor delay, ages 15 yrs at follow up	MABC Various movement tasks	19 perceptual and motor tasks Physical activity interview	Performance on all motor tasks of DCD group poorer than that of controls, and differences still existed 10 yrs later	DCD group believed themselves to be less physically competent, and had fewer physical spare-time activities
7	Cantell (2008)	Cross-sectional	110 children/adults with high or low motor competence, ages: 8–9 yrs, 17–18 yrs, and 20–60 yrs	MABC DCDQ	Body composition CRF Flexibility Muscle strength Lung capacity Leisure participation	Low motor competence group had higher BMI scores, greater percentage body fat, and poorer fitness results in endurance, flexibility, and strength.	DCD children (females) spent less time in mild, moderate, and strenuous activity
8	Castelli (2007)	Cross-sectional	230 children, with low and high motor competence, ages 7-12 yrs	SCPEAP, Motor task performance	Body composition CRF Muscle strength Flexibility Physical activity (recall and accelerometer)	No correlation between motor competence and BMI, flexibility. Inverse correlations between motor competence and aerobic fitness, muscle strength	Motor competence was a predictor of physical activity
9	Causgrove Dunn (2006)	Cross-sectional	130 children with and without movement difficulties, ages 9-12 yrs	TOMI	Physical activity participation during PE class	NA	DCD group spent less time successfully engaged in assigned activities and spent more time engaged in off-task behaviors. Motivational variables were important.
10	Chia (2009)	Cross-sectional	31 boys with and without DCD, ages 7-10 yrs	MAND	Maximal CRF (VO2 Peak)	DCD group achieved lower VO2 peak relative to comparison group	NA

11	Christiansen (2000)	Cross-sectional	30 boys with and without deficits in attention, motor control and perception, ages 11-12 yrs	MABC	Spare time sport activities	NA	DCD group avoided participation in team sports compared to controls
12	Fisher (2005)	Cross-sectional	394 children with low to high movement skill scores, ages 3-5 yrs	MABC	Physical activity (accelerometer)	NA	Total movement skills score was weakly but significantly positively correlated with total physical activity accelerometry output
13	Haga** (2008a, 2008b)	Cross-sectional	67 children with low and high motor competence, ages 9-10 yrs	MABC	CRF Muscle strength Power Anaerobic capacity	DCD group performed worse for all nine tasks on the test of physical fitness relative to the comparison group	NA
14	Haga (2009)	Longitudinal (32 months follow up)	67 children with low or high motor competence, ages 9-10 yrs	MABC	Anaerobic capacity CRF Power Muscle strength	DCD group performed consistently lower on all physical fitness measures over time	NA
15	Hands (2006)	Cross-sectional	104 children with and without motor learning difficulties, ages 5-8 yrs	MAND MABC	Body composition CRF Muscle strength Power Anaerobic capacity Flexibility	DCD group had higher BMI and lower performance levels on the sit and reach, sit-ups, standing broad jump, 50-meter run, and the shuttle run	NA
16	Hands (2008)	Longitudinal (5 yrs follow up)	38 children with high and low motor competence, ages 5-7 yrs at baseline	SIS	Body composition CRF Anaerobic capacity Power Muscle strength	DCD group performed consistently lower on physical fitness measures: jumping, 50-m run, throws, and cardio-respiratory endurance. There were no differences for BMI. Aerobic fitness differences worsened over time	NA

17	Hands (2009)	Cross-sectional	1585 children with high and low motor competence, age 14 yrs	MAND	Body composition CRF Muscle strength Flexibility Physical activity (pedometer)	DCD group had poorer performance on all measures	No differences in physical activity between groups as measured by pedometers
18	Hay (1998)	Cross-sectional	492 children with high and low self-efficacy and motor proficiency, ages 10-14 yrs	BOTMP	Physical activity participation	NA	Children with poor adequacy and predilection for physical activity were found to be less motorically competent, and were less physically active in both free and organized play
19	Hay (2007)	Longitudinal (24 months follow up)	1282 children with high and low motor proficiency, ages 10-11 yrs	BOTMP	Body composition	BMI and waist girth increased more rapidly in children with poorer motor proficiency	NA
20	Kanioglou (2006)	Cross-sectional	154 children with and without DCD, mean age 10.9 yrs	MABC	CRF Anaerobic capacity Power Muscle strength	DCD group had poorer performance in 50-yard sprint, 600-yard run, shuttle run, sit-ups, and long jump. Muscle strength was lower, but not statistically significant	NA
21	Kaufman (2007)	Longitudinal Case study	1 boy with DCD, age 5 yrs	BOTMP	Body composition Muscle strength	Child was obese, had poor muscle tone, decreased endurance, hyper-extensibility. Muscle strength showed improvement following 12 week strength training program	NA
22	Mata (2007)	Cross-sectional	221 children with high and low motor competence, ages 12-14 yrs	BCTC	CRF	DCD group had lower peak VO2 values as measured by the 20m shuttle run	NA

23	Mandich (2003)	Cross-sectional	12 parents of children with DCD (10 children total ages 7-12 yrs)	DSM-IV	Interviews regarding participation in activities	NA	Children with DCD experienced activity limitations and restricted participation both in terms of motor skills and social consequences according to parent interviews
24	O'beirne (1994)	Cross-sectional	48 boys with poor and normal coordination, ages 7-9 yrs	MAND	Body composition Anaerobic capacity	Poorly coordinated group was heavier, and had lower scores on the Wingate anaerobic test and the 50 m sprint	NA
25	Okely (2001)	Cross-sectional	1844 children with various movement skills, ages 13-15 yrs	Movement skills assessment	Physical activity recall	NA	Fundamental movement skills predicted time in organized physical activity, but the percentage of variance explained was small. Prediction was stronger for girls
26	Peters (1999)	Longitudinal (10 weeks follow up)	14 children with DCD, ages 7-8 yrs	MABC DSM-IV	Muscle strength	Children showed low muscle tone/joint hyper-extensibility. Forced muscle capacity increased following 10 week exercise intervention	NA
27	Poulsen** (2006; 2007a,b; 2008a,b)	Cross-sectional	173 boys with and without DCD, ages 10-13 yrs	MABC	Recall of leisure-time behaviour	NA	DCD group recorded lower participation rates in all group physical activities, whether structured (e.g., team sports) or unstructured (e.g., informal outdoor play) and lower energy expenditure
28	Raynor (2001)	Cross-sectional	40 children with and without DCD, ages 6-10 yrs	MAND	Muscle strength Power	DCD group showed decreased power, and it was more apparent at higher velocities of movement, as well as a lower flexor-extensor percentage was recorded for DCD group	NA
29	Reeves (1999)	Cross-sectional	51 children with various motor skills, ages 5-6 yrs	BOTMP	CRF	Negative correlation between ½ mile performance and motor skills	NA

30	Schott (2007)	Cross-sectional	261 children with and without DCD, ages 4-12 yrs	MABC	Body composition CRF Anaerobic capacity Muscular strength Power Flexibility Physical activity recall	More overweight/ obese children with DCD in the 10-12yrs old group. DCD group performed worse in the 20m sprint, 6 min run, jump-and-reach test, and ball throw, but not flexibility	8.5-15% of the DCD groups engaged in adequate physical activity i.e. 2hr/day (severe and moderate, respectively) vs. 19-21% of typically developing children (medium and high MABC respectively)
31	Silman (2010)	Cross-sectional	122 children with and without DCD, ages 12-13 yrs	MABC	Body composition Maximal CRF (VO2 peak) Physical activity (accelerometer)	DCD group had greater percentage of body fat and lower peak VO2.	DCD group was significantly less active during the 7 day monitoring period using accelerometers.
32	Smyth (2000)	Cross-sectional	110 children with and without DCD, ages 6-10 yrs	MABC	School playground observations	NA	DCD group spent more time alone, played games in large groups less often, and some tended not to get involved in social physical play
33	Smyth (2001)	Cross-sectional	64 boys with and without DCD, ages 6-10 yrs	MABC	School playground observations	NA	DCD group spent more time alone. Only poor balance score on MABC subscale negatively affected football participation
34	Tsiotra (2006)	Cross-sectional	591 Canadian and 329 Greek children with and without DCD, mean age=11.46 and 11.3 respectively	BOTMP	Body composition CRF	Greater prevalence rates for obesity (% body fat) and lower aerobic fitness (shuttle run) observed in the DCD group relative to non DCD and in the Greek sample relative to Canadian sample	NA
35	Tsiotra (2009)	Cross-sectional	177 Greek children with and without DCD, ages 10-12 yrs	BOTMP	Body composition CRF Muscle strength Flexibility Power Anaerobic capacity	Although DCD children had lower values in all six fitness parameters, only four (i.e., BMI, power, hand strength, and 40m speed test) were found to be significantly different	NA

36	Ulrich (1987)	Cross-sectional	250 children with various motor competence skills, ages 5-10 yrs	Nine item motor competence battery	Sport participation	NA	Participation in organized sports was positively related to motor competence, but perceived competence was not
37	Visser (1998)	Longitudinal (30 months follow up)	30 boys with various motor competence skills, ages 11 yrs at baseline	MABC	Physical activity recall	NA	Mean amount of physical activity was consistently lower in the DCD group, but the gap diminished over time
38	Williams (2008)	Cross-sectional	198 children with various motor skills, ages 3-4 yrs	CMSP	Body composition Physical activity (accelerometer)	No correlation between BMI and motor skills score	Children with poorer motor skill performance were less active, but the effect size was small
39	Wrotniak (2006)	Cross-sectional	65 children with high and low motor proficiency, ages 8-10 yrs	BOTMP	Body composition Physical activity (accelerometer)	Poor motor proficiency was correlated with greater BMI scores	In the highest quartile of BOTMP, motor proficiency was positively associated with activity counts and percentage of time in moderate and vigorous intensity physical activity, no difference for those in the lower 3 quartiles
40	Wu (2010)	Cross-sectional	41 children with and without DCD, ages 9-11 yrs	MABC	Body composition CRF	No significant differences between groups in BMI. DCD group had lower peak VO2 results and ran 800m in a slower time than the typically developing children	NA

CRF=Cardiorespiratory fitness.

SCPEAP=South Carolina Physical Education Assessment Program.

BCTC=Body Coordination Test for Children.

SIS=Stay in step -a gross motor screening test.

CMSP=Children's Activity and Movement in Preschool Study Motor Skill Protocol.

*DCD used as a generic term, and may refer to low motor competence, movement difficulties, etc. as defined by each study.

**Multiple publications utilizing the same study population.

2.4. Summary of Study Results & Discussion

The purpose of this study was to systematically review and summarize the literature on the association between poor motor proficiency and fitness and physical activity outcomes in children. Two clear findings emerged from this systematic review. The first is that children with poor motor proficiency generally had poorer performance than their peers on most measures of physical fitness. Second, these children were less physically active than their peers.

2.4.1. Body composition

Of the 18 studies that assessed the effect of motor proficiency on body composition, the majority (n=13) reported that children with poor motor proficiency had greater weight, higher BMI scores, greater waist girth, and greater percentage body fat relative to their peers. Differences between study groups were significant in 10 of the 15 studies that assessed BMI (Cairney et al., 2005-2010; Cantell et al., 2008; Faught et al., 2005; Hands & Larkin, 2006, Hands et al., 2009; Hay et al., 2007; Kaufman & Schilling, 2007; O'Beirne et al., 1994; Schott et al., 2007; Tsiotra et al., 2009; Wrotniak et al., 2006), the most common anthropometric measure used in the reviewed studies. Other less commonly used measures included assessments of body fat percentage (e.g., whole body air-displacement plethysmography, bioelectrical impedance), as well as indirect measures such as waist circumference (WC), and weight in kg.

Using BMI as a measure, Cairney et al. (2005a) found that DCD was a risk factor for overweight and obesity. Specifically, the prevalence of overweight and obese was 19.2% greater in boys with DCD compared to the typically developing children, although

for girls there were no differences attributable to the disorder. Cairney et al. (2010a) observed a similar trend in a longitudinal study, where BMI differed by approximately 15%, with the DCD group being heavier at baseline. Furthermore, the gap between DCD and typically developing children remained stable over time. Cantell et al. (2008) showed a similar difference in prevalence, with 52% of the low motor competence group being overweight or obese compared to 30% of the high motor competence group. Once the analysis was stratified by gender, this effect was only significant for females ($p = .087$). Hands & Larkin (2006) reported that 15.4% of children with motor learning difficulties had BMI over 20, versus only 5.8% of controls. In a sample of 7-9 yr old boys, O'Beirne et al. (1994) observed that the poorly coordinated children were on average 15% heavier. Negative correlations between BOTMP and BMI have been reported in recent studies (Hay et al., 2007; Wrotniak et al., 2006). Consistent with the above findings, WC showed a similar trend. Hay et al. (2007) observed -0.33 correlation between BOTMP and WC, while Cairney and colleagues (2010a) reported a WC difference of 12% between DCD and non DCD groups at baseline.

Studies that measured body fat percentage found significant associations with DCD status. Silman et al. (in press) found that the DCD group had 40 percent greater body fat percentage than the controls, using whole body air-displacement plethysmography. Cairney et al. (2005a), reported that children with DCD were more likely to be overweight and obese (23.3%) than children without the disorder (12.1%) when percentage body fat was examined in a bivariate analysis. However, there were no significant differences in overweight and obesity between children with DCD and healthy controls when BMI was used in the same analysis. In an international comparative

investigation between Canadian and Greek children, Tsiotra et al. (2006) demonstrated that Greek children with DCD had a 23% greater prevalence of obesity than their non DCD counterparts, as measured by bioelectrical impedance analysis. In contrast, the Canadian sample in the same study, which was overall less heavy, the prevalence of obesity was 11% higher among the DCD children. It was also noted that the Greek sample was relatively inactive compared with their peers from other countries and generally showed greater prevalence of obesity and overweight than Canadian children, irrespective of DCD status, potentially accounting for the higher prevalence rates of obesity in that sample.

The effect of gender on the association between motor competence and body composition was examined in a small number of cross-sectional studies demonstrating varying results. Schott et al. (2007) found significantly greater BMI in boys with severe DCD relative to girls. Likewise, Cairney et al. (2005a) reported that DCD was a risk factor for overweight and obesity in boys. While the propensity for overweight and obesity was even greater in girls, the overall risk could not be attributed to DCD. Conversely, Wrotniak et al. (2006) showed that, although poor motor proficiency was correlated with greater BMI scores, there was no gender effect. Considering longitudinal effects, two of the three studies that examined the relationship between DCD and body composition over time have demonstrated that differences in both BMI and waist circumference remained significant over time, and even increased more rapidly in children with poorer motor proficiency, (Cairney et al., 2010a; Hay et al., 2007). Where gender was considered, the trajectories were similar for both boys and girls, regardless of DCD status, throughout the follow up period (Cairney et al., 2010a).

Among the few negative studies, Hands (2008) found neither a main effect for motor coordination on BMI, nor a time by motor coordination interaction, in her analysis of 5 to 7 year old children. Similarly, Schott et al. (2007) did not find an association between DCD and BMI in 4-9 year old children, although in the older group (10-12 yrs old) the percentage of overweight and obese children in the DCD group was significantly higher. Furthermore, no correlation between BMI and motor skills score was found by Williams et al. (2008) in a sample of 3-4 year old children. These discrepancies between studies may be due to the relatively small number of participants (n=38) in Hand's study, or differences in the age composition of the samples. In fact, Hands & Larkin (2006) study, utilizing a larger sample size (n=104) it was found that the group with motor learning difficulties did in fact have a significantly higher BMI. It is arguable that the detrimental effect of poor coordination on body composition does not manifest itself until later in childhood, early adolescence. Therefore, body composition may not be significantly associated with DCD in younger cohorts. Moreover, in early childhood as children become more engaged in physical activities and organized sports, those with poor motor coordination are potentially at a greater risk for inactivity because of their disorder, and therefore are more prone to weight gain and obesity.

Measurement issues may potentially play a role in obscuring the relationship between body composition and motor competence. Specifically, BMI has been reported to have poor sensitivity in screening for overweight children (Mast et al., 2002). For example, in a sample of 578 children, Cairney et al. (2005a) noted that while there was acceptable agreement between the body composition measures, using BMI classification resulted in a total of 90 children who meet the criteria for overweight or obesity

compared with 75 children using percentage body fat as the outcome measure. It is recommended that in future studies body composition analysis be used to screen for children at risk of becoming obese as BMI may not be a sensitive enough measure.

The overwhelming evidence supports an increased risk for elevated body fat in children with DCD. Several mechanisms can contribute to the observed effects. Children with DCD demonstrate decreased levels of participation in physical education, organized play, and general physical activity (Bouffard et al., 1996; Cantell et al., 2008; Christiansen, 2000). Decreased self-efficacy toward physical activity (Cairney et al., 2005b) may also result in activity avoidance. This hypoactivity may lead to an energy imbalance whereby energy expended is less than energy consumed, leading to an accumulation of body fat (Tsiotra et al., 2009). Furthermore, an increase in BMI and body fat may directly affect children's performance on activities such as running, jumping, and flexibility, independent of the effect of DCD, due to the mechanical disadvantage of excess weight and the higher oxygen cost of locomotion (Hands & Larkin, 2002). This same relationship exists in the normally coordinated population of children, but the impact is clearly more profound over time in children with poor coordination (Cairney et al., 2010a).

2.4.2. Cardiorespiratory Fitness

A total of 19 studies provided evidence on the relationship between poor motor proficiency and cardiorespiratory fitness. A number of protocols exist for estimating aerobic fitness. These can be divided into laboratory-based methods that evaluate aerobic power or $\text{VO}_{2\text{peak}}$ (maximal volume of oxygen consumed), which tend to be resource

and time-intensive, but highly accurate; and field-based methods, which estimate VO_2peak indirectly based on measures of physical performance and are practical in the absence of specialized equipment (Cairney et al., 2010b). The measure most commonly used to evaluate cardiorespiratory fitness was the Léger 20-m shuttle run assessment (Léger & Lambert, 1982), which was performed in 11 studies. Direct laboratory assessments of maximal aerobic capacity measuring VO_2max were utilized in four studies. Of these, two were performed using an incremental treadmill protocol, while the remaining two studies were conducted on a bicycle ergometer. Other assessments of aerobic fitness included the 6-min run and half-mile run, where the authors compared the distance covered on the test by the DCD relative to the non-DCD children. Overwhelmingly, 18 of these studies reported that children with DCD demonstrated lower aerobic power compared to their typically developing peers. Tsiotra et al. (2009) was the only study that failed to demonstrate a relationship between DCD and aerobic fitness in their sample of Greek, 10-12 year old children. It was hypothesized that the results may be attributed in part to the fact that Greek children generally exhibit lower aerobic fitness compared with children from other countries (Bouziotas et al., 2001), thus making it difficult to discern the effect of DCD.

The magnitude of the effect size in cardiorespiratory fitness varied between studies, and was also reported differently among studies, making direct comparisons challenging. For example, Cairney et al. (2007) reported that DCD is associated with lower cardiorespiratory fitness across all age groups (9–14 yrs), showing that VO_2max , as estimated from the shuttle run, was on average 17% lower in the DCD group relative to the comparison group (31.4 ml/min/kg vs. 38.0 ml/min/kg, respectively). Moreover, it

was found that children with DCD were much more likely (61%) than their non-DCD peers (23%) to have predicted VO₂max scores in the bottom 20th percentile. Mata et al. (2007) reported a significant difference in cardiorespiratory fitness between children with DCD and those without; however, the magnitude of the difference was not provided. Castelli and Valley (2007) showed a significant correlation ($r=0.57$) between performance on the shuttle run and motor competence scores. Another study using the Léger shuttle run, found that children with motor learning difficulties ran significantly fewer laps during the test $n=11$ vs. $n=15$, respectively; a difference of 27% (Hands & Larkin, 2006). Using similar testing procedures, Kanioglou (2006) reported large effect sizes between the control group and children with moderate motor difficulties and severe motor difficulties (eta squared=0.41 and 0.48, respectively). Hands (2008) demonstrated a similar effect size (eta squared=0.38) comparing groups of children with low and high motor competence. Tsiotra et al. (2006) compared Canadian and Greek children on the shuttle run, showing that in both populations, children with DCD demonstrated poorer cardiorespiratory fitness profiles. Specifically, the prevalence rate for low cardiorespiratory fitness in the Canadian sample was 83% in the DCD group and 55% in the non-DCD group, while in the Greek sample the rates were 90% vs. 65% in the DCD and non-DCD groups, respectively.

Three studies used the six-minute run to compare children's aerobic fitness, all showing modest effect sizes. For example, a significant difference was demonstrated in Haga (2008a), whereby children in the high motor competence group covered on average 10% greater distance on the six-minute run test (986m) compared to the low motor competence group (895m) ($p=0.05$). In a recent longitudinal study, Haga (2009) reported

a significant difference between children with high and low motor competence, as those with motor difficulties covered 11% less distance at baseline. This difference continued to exist between the groups after a 2 year follow-up, whereby the group's performance was on average 13% lower. Similarly, Schott et al. (2007) demonstrated modest differences between children with DCD (severe and moderate) and typically developing children (medium and high motor competence), with the actual distance covered in 6 minutes ranging from 797m to 929m between the four groups, with the severe DCD group having the poorest outcome.

The four studies that measured aerobic power in a laboratory setting showed significant differences between motor competence groups, and effect sizes were similar to field based assessments. Specifically, VO_{2peak} was 17% lower in the DCD group compared to the non DCD group in Wu et al. (2009) (39.7 vs. 47.6 ml/kg/min, respectively). Similarly, Silman et al. (in press) found an 18% difference between the DCD and non DCD groups in her sample (35.0 vs. 42.9 ml/kg/min, respectively). Hands et al. (2009) reported that the high motor competence group had an 11% greater physical capacity on the PWC 170 test. This test indirectly provides an estimate of VO_{2max} by extrapolating the load required for a heart rate of 170 and is considered a suitable measure of aerobic fitness for this age group (Rowland et al., 1993). A 22% difference in VO_{2max} was reported in Chia et al. (2009) using an incremental treadmill protocol.

Three longitudinal studies have examined the changes in cardiorespiratory fitness over time, demonstrating that the negative effects of poor motor proficiency persist as children grow older (Cairney et al., 2010b; Haga, 2009; Hands et al., 2008). Hands (2008) showed that children with low motor competence never caught up to the high

motor competence group in their performance on the shuttle run over a five year period. In fact, their performance worsened over time, reporting a large effect size (partial eta squared = 0.38). Likewise, Cairney et al. (2010b) reported that not only was the difference in VO_2 peak between children with and without DCD significant at baseline in a sample of 2278 children, it declined at a much greater rate in the DCD group, suggesting that the difference in cardiovascular endurance persists and diminishes more rapidly over time. Specifically, in healthy boys, VO_2 peak ranged from 48.5 at baseline (grade 4) to 48.0 at follow up (grade 6), for girls the range was 46.6 to 46.0 over the same time period. While in the DCD group VO_2 peak ranged from 43.2-42.1 in boys and 43.6-41.7 in girls. Haga (2009) also found that children with low motor competence had consistently lower aerobic fitness results over a 32 month follow-up period, although no interaction effect with time was evident, potentially due to the small sample size ($n=67$).

A concern with field-based measures of aerobic capacity is that they rely on the internal motivation of the child to perform to exhaustion. This is particularly challenging for children with DCD as they generally perceive themselves to be less competent and may have less motivation to continue the assessment potentially dropping out prematurely, underestimating the true aerobic capacity of this group (Cairney et al., 2006a; Hay et al., 2007; Silman et al., in press). However, in the laboratory setting, other indicators such as heart rate (HR) and respiratory exchange ratio (RER) can more accurately monitor a child's performance to ensure a true VO_2 peak is achieved (Silman et al., in press). A closer examination of these two techniques in determining VO_2 peak is valuable considering that the laboratory method would allow children to feel more comfortable to perform the assessment without feeling self-conscious around their peers

while receiving positive feedback from the lab technician. This was demonstrated by Silman et al. (in press), whereby children with DCD achieved comparable maximum HR and RER measures to their non-DCD peers, indicating that the subjects who were poorly coordinated were working as hard as the typically developing subjects. The author also highlighted the importance of adequate motivational encouragement in order to assist the child; especially those children with DCD, to achieve maximum effort when performing a VO₂peak test. Cairney et al. (2010c) addressed the issue of comparability of the shuttle run and the lab based cycle ergometer tests for assessing cardiorespiratory fitness, showing moderate to good correlations between the two tests. Nonetheless, the findings of the laboratory based studies corroborate those reported in field tests, suggesting a true deficit exists and that the effect of DCD on aerobic fitness is not limited by the influence of perceived competence.

2.4.3. Muscle Strength, Endurance and Flexibility

All 14 of the reviewed studies that examined muscle strength and endurance reported a negative effect of low motor proficiency on this fitness parameter. Studies that utilized comparison groups showed that the low motor competence group had significantly poorer performance relative to control subjects on several fitness indices including: number of sit-ups, push-ups performed in a specified amount of time, hand grip force, and ball throw distance. Effect sizes relative to gender, age and type of assessment varied. For example Cantell et al. (2008) examined muscular endurance by assessing the number of curl-ups performed in 30 seconds, stratified by age and gender. In the 8-9 yrs group, boys in the high motor competence group outperformed those with

low motor competence by 46%, whereas females of the same age group with high motor competence outperformed their peers by 60%. In the older age group (17-18 yrs), boys continued to perform better, albeit a lesser degree (15%), while no difference was observed for the girls. Kanioglou (2006) observed that children with adequate motor skills completed significantly more sit ups in 60 seconds than children with moderate (25%) and severe motor difficulties (37%) (eta squared 0.68 and 0.49, respectively). Haga (2009) found that children with low motor competence had consistently lower muscle strength as measured by the medicine ball throw. The high motor competence group had on average 21% greater throwing distance at baseline and 20% greater distance when assessed 32 months later at follow up.

Six of the reviewed studies examined the effect of low motor proficiency on flexibility and those provided mixed results. Three studies reported poorer flexibility on the sit and reach test for the low motor proficiency group (Cantell et al., 2008; Hands & Larkin, 2006; Hands et al., 2009), while Schott et al. (2007), Castelli and Valley (2007), and Tsiotra et al. (2009) did not find this relationship. Specifically, Cantell et al. (2008) reported that the low motor competence group scored significantly lower than the high motor competence group; partial eta squared=0.088. Hands & Larkin (2006) reported an R squared of 16.8% using a generalized linear model analysis (adjusted for gender), while in a more recent study Hands et al. (2009) found significant correlations between flexibility and motor competence score, $r=0.22$ for females and $r=0.26$ for males. According to Hands (2008), children with poor motor proficiency tend to have heterogeneous fitness profiles, which may result in extreme ranges of flexibility or rigidity. O'Beirne & Larkin (1991) (in Cermak & Larkin, 2002), reported that 73% of

children with DCD scored above the 75th percentile or below the 25th percentile on the sit and reach test, demonstrating that the range of motion of this group of children varies dramatically.

Adequate muscle strength and endurance are important for performing many daily activities and sports without fatigue. Poor muscular strength may result in poor posture, musculoskeletal problems such as lower back pain, lax joints, and difficulty participating in sports, particularly those requiring production of force (Hands & Larkin, 2002). Children with DCD may withdraw from physical activities that require continued use of muscle groups due to poor endurance and early fatigue, which in turn will hinder the development of both muscle strength and endurance. Moreover, excessive flexibility may result in joint instability, making it difficult to perform controlled movements, while lack of flexibility may result in inability to perform movements efficiently (Hands & Larkin, 2002).

2.4.4. Anaerobic Capacity

All eight studies that examined anaerobic capacity have shown that subjects with poor motor proficiency had lower anaerobic performance compared to typically developing, or highly motorically proficient children. Anaerobic performance was typically measured by running 20 or 50 meters at maximal speed. O'Beirne et al. (1994) used both the 50 meter run and the Wingate cycle ergometer test to assess anaerobic performance, which required subjects to pedal at maximum speed for 30 seconds. Relative peak and mean power using the Wingate test correlated with the MAND with values of $r = 0.59$ and $r = 0.74$, respectively. For the 50 meter run test, O'Beirne et al.

(1994) analyzed the results stratified by age, to show that differences between groups increased with age. Specifically, there was a difference between the motor proficiency groups of 16% for the 7 year olds, 19% for the 8 year olds, and 25% for the 9 year olds. Other studies reported small to moderate effect sizes in anaerobic performance, with observed differences between groups of 7 to 30%. The smallest difference (7%), although statistically significant, was reported in Haga (2008a) in 9-10 yrs old sample of high and low motor competent children on the 20 m sprint. The largest difference was reported by Hands (2008), who observed a difference of 30% between high and low motor competent children at baseline, although only a 15% difference was observed at follow up 5 yrs later.

Similarly, all seven studies that have compared the performance on measures of explosive power have found that those with normal or high motor proficiency consistently outperformed those with low motor proficiency. Measures of anaerobic power typically included the standing broad jump, vertical jump, and throwing a medicine ball. Differences between motor competent children and those with poor motor proficiency varied between 10%-30% in the reviewed studies. The smallest effect size was reported in Kanioglou (2006), who found that typically developing children covered 10% and 15% more distance on the standing broad jump, than those with moderate and severe DCD, respectively. Hands and Larkin (2006) reported the largest effect size using the same measure (standing broad jump), with 30% difference between children with and without motor learning difficulties. In addition to the overall group differences in running speed, O'Beirne et al. (1994) reported that in their sample of 7-9 yr old subjects, those who were poorly coordinated demonstrated less age related

improvement. The authors also found a correlation between age and power output, however power output in the low motor proficiency group did not increase, which may explain why no improvement in anaerobic performance was observed. Studies that utilized a longitudinal study design confirmed that while all children generally improved over time in both anaerobic performance and power, the improvement of the low motor proficiency group was consistently less over time. In a 5 year follow up study, Hands (2008) observed time by group interactions on the standing broad jump, indicating that the low motor competency group's performance worsened with time relative to the high motor competency group (23% difference at baseline and 18% at follow up).

Overall, these studies suggest that children who perform poorly on motor skills have poor anaerobic performance and power, and that they are unlikely to catch up to their peers with age. Hands and Larkin (2006) found that performance on the standing broad jump had the highest variance explained by motor competence status, explaining more of the variance between the groups than any of the other measures of fitness (i.e., BMI, sit and reach, sit-ups, grip strength, chest pass, standing broad jump, 50 meter run, shuttle run). Jumping requires good coordination and dynamic balance to achieve optimum performance. Since children with poor motor proficiency generally lack these abilities, it is not unexpected that performance on power assessments was also deficient (Hands & Larkin, 2006). It has also been suggested that poor performance on anaerobic tasks may be explained by deficient neuromotor control and motor fiber recruitment (Keller et al., 2000; O'Beirne et al., 1994). Furthermore, increased muscle fatigue, which may reflect mechanically inefficient movement patterns in the low motor proficient children, may also contribute to a reduction in anaerobic performance, as confirmed in

O'Beirne et al. (1994) who observed that subjects who were poorly coordinated were not able to maintain as great percentage of power output and reported greater local muscle fatigue.

2.4.5. Physical Activity

Poor motor proficiency was associated with lower levels of physical activity and participation in free and organized play in 20 of the 21 studies that examined these outcomes. Physical activity was measured using various instruments, most commonly questionnaires that relied on participants' recall (n=10 of the reviewed studies). Four studies utilized direct measurements of physical activity including the use of accelerometers (Castelli & Valley, 2007; Fisher et al., 2005; Silman et al., in press) and a pedometer (Hands et al., 2009). Four studies relied on observations of children either at the school playground during recess, or by observing children's involvement in physical education classes (Bouffard et al., 1996; Causgrove Dunn & Dunn, 2006; Smyth & Anderson, 2000, 2001). Three studies utilized semi-structured interviews, some directly with the children (Cantell et al., 1994; Christiansen, 2000), or in the case of Mandich et al. (2003), interviewed parents of children with DCD to get an in-depth understanding of participation in daily activities and the impact on the lives of families coping with DCD.

The measures used in the reviewed studies differed in their operationalization of the construct of 'physical activity', making direct comparisons between them difficult. Some (Visser et al., 1998; Ulrich, 1987) noting the lack of well-developed assessment instruments, chose to construct their own questionnaires to measure participation in physical activity without formally validating the instruments. For example, Ulrich (1987)

assessed whether or not each subject participated in at least one organized sport in the past year and classified children as either participants or non-participants for analysis. On the other hand, Hay and Missiuna (1998), Cairney et al. (2005-2007), and Faught et al. (2005) used a more comprehensive tool, the Participation Questionnaire (PQ). The PQ is a 61-item questionnaire that asks children to report on their participation levels in the areas of free-time play, seasonal recreation pursuits, and various sporting activities. It has been demonstrated to have strong construct validity and good test-retest reliability (Hay, 1992; Hay, 1999). Cantell et al. (2008) used another subjective instrument; the Godin Leisure-Time Exercise Questionnaire (Godin & Shephard, 1985). This tool, which has been previously validated (Salis et al., 1993), has participants record the amount of time spent on three different levels of physical activities during the previous seven days. The authors then calculated energy expenditure in metabolic equivalents and categorized children on a scale of physical activity from 1='needs improvement' to 5='excellent' for their analysis. Other measurement tools included various means relying on participants' recall of physical activity in the past seven days (e.g., Castelli & Valley, 2007; Okely et al., 2001; Schott et al., 2007). Barnett et al. (2009) chose to adopt the Australian Physical Activity Recall Questionnaire, which assessed the type of activity, frequency, and duration of physical activity, and has been validated in Booth et al. (2002).

Studies utilizing self reported measures of physical activity varied in their effect sizes. For example, Cairney et al. (2006b) found that children with DCD participated significantly less in organized and free play than their non DCD peers, but observed a small effect size, partial eta squared = 0.012 and 0.010, respectively. Conversely, Cantell et al. (2008) found a large effect size reporting that the low motor competence group had

significantly lower scores on the Leisure Score Index (Godin & Shephard, 1985) than the high motor competence group, partial eta squared = 0.693. However, this relationship was only observed for children (8-9 yrs), and no significant association emerged in adolescents (17-18 yrs). Hay and Missiuna (1998) observed that children with poor adequacy and predilection for physical activity were found to be less motorically competent, and were less physically active in both free and organized play, PQ total and BOTMP were significantly correlated ($r = 0.57$), and the relationship grew stronger with age. Poulsen et al. (2008b) measured metabolic (MET) levels from energy expenditure as calculated from a 7-day leisure time diary to show that significant differences exist between those with and without DCD (partial eta square = 0.25 for total MET score between groups). Schott et al. (2007) reported that 8.5-15% of the DCD group engaged in adequate physical activity (severe and moderate, respectively) vs. 19-21% of typically developing children (medium and high MABC, respectively). It should be noted that sufficient physical activity level was defined as spending at least 60 minutes a day in moderate-to-vigorous activities. Regarding exercise intensity, an interesting finding reported by Cantell et al. (2008) was that there were significant differences between children in the low and high motor competence groups. Moreover, there was a trend for a higher sweat score in individuals with low motor competence suggesting that they were more taxed and less efficient during exercise than individuals with high motor competence.

Most studies relying on more objective measures of activity such as pedometers and accelerometers reported smaller effect sizes, with the exception of Castelli and Valley (2007) who found a significant correlation between total motor competency score

and steps taken during formal activity program instruction as measured by a pedometer ($r = .54$). Wrotniak et al. (2006) categorized children according to BOTMP scores and showed that those in the highest quartile had significantly greater average activity and percentage of time spent in moderate-vigorous physical activity compared with those in the lower BOTMP quartiles as measured by accelerometers. However, no differences in physical activity among children in the lower three quartiles were evident. It was also found that motor proficiency explained an additional 8.7% of the variance in physical activity after controlling for child gender, socioeconomic status, televisions in the home, children in the home, child's BMI, one parent's BMI, and CSAPPA score. This is larger than the 3% of the variance in time spent in organized physical activity explained by movement skills reported previously by Okely et al. (2001). The authors speculate that the lower value reported by Okely and colleagues (2001) may be the result of underestimation due to physical activity being self-reported rather than objectively measured, a limited range of movement skills being tested, and the difference in ages of the children. Williams et al. (2008), using an accelerometer, found that children with poorer motor skills were less active although the effect size was also small. For the total group, there was a statistically significant relationship between total motor performance scores and physical activity ($r = 0.20$ for moderate-vigorous activity and $r = 0.26$ for vigorous activity). Finally, Fisher et al. (2005) reported total physical activity and percent time spent in moderate to vigorous physical activity (accelerometry output) were significantly but weakly correlated with total movement skills score ($r = .18$ for moderate-vigorous activity). The correlations were very similar for both boys and girls. It should be noted that use of accelerometry to measure physical activity offers many improvements over

self-report techniques; however this method is not without its challenges. There is currently no clear consensus on scoring and interpretation of accelerometry data to measure physical activity behaviour (Ward et al., 2005).

Several studies utilized direct observational methods to assess participation in physical activity. Bouffard et al. (1996) found significant differences between children with and without movement difficulties in the amount of vigorous activity during recess. Control subjects were vigorously active for 23.7% of the time compared to subjects with movement difficulties who were only vigorously active 15.1% of the time. No differences between regular activity and inactivity were noted. Causgrove Dunn and Dunn (2006) observed a similar effect size, it was reported that during physical education classes, children with motor difficulties spent an average of 5.72% less time than their matched classmates engaged in adaptive behaviors and 5.44% more time engaged in maladaptive behaviors (e.g., assuming non-participant roles). While the size of these mean difference scores are small in terms of absolute time spent in activities in one class; the cumulative effect of these differences over time may result in substantial inactivity (Bouffard et al., 1996; Wall, 2004). Smyth and Anderson (2000) observed children's school playground activity, and found that children with DCD participated in significantly less formal and informal team games, spent more time alone, and were onlookers more often than those without DCD.

A total of four longitudinal studies assessed participation in physical activity. Barnett's et al., (2009) results suggest that being able to perform object control skills (e.g. skills involving manipulation of an object such as a ball) competently in childhood may be a significant factor in predicting subsequent engagement in physical activity during

adolescence. Predicted values showed that children with good object control skills have at least a 20% greater chance of participating in some vigorous activity in adolescence (6-7 years later), compared with those with poor object control skills. Cantell et al. (1994) reported that at age 15, the group diagnosed with having delayed motor development reported significantly fewer spare-time activities (e.g., social and physical hobbies). The mean number of hobbies was 1.75 at age 15 in the group with motor problems versus 2.85 in the control group. This study did not measure the same outcomes at baseline and therefore it was not possible to assess how the difference between groups changed over time. Similarly, Cairney et al. (2006b) found no evidence to support the divergence in activity-deficit with age hypothesis using a sample with a broader age range of children (9 to 14 yrs) when participation in both structured and unstructured play opportunities are considered. Furthermore, even though the outcomes in both these studies were different (self-reported versus observational measures), the results were congruent. The mean amount of physical activity was consistently lower in the DCD group, but the gap diminished over time (Visser et al., 1998). Specifically, at age 11 yrs, the DCD group averaged half the amount physical activity per week (4 hrs) compared to the non DCD group (8 hrs). This trend continued with age (12 to 14 yrs) despite an increase in physical activity per week in the DCD (5 hrs) and non DCD (7.5 hrs). When considered together, the results of these longitudinal studies suggest that, although children with DCD seem to be less likely to participate in free play or organized activities, the deficit does not increase with age.

Hands et al. (2009) was not able to demonstrate the negative association between poor motor proficiency and physical activity reported in other studies. In this study,

physical activity was assessed by a pedometer and therefore it was not possible to assess the intensity, type, or frequency of the activity. While pedometers record locomotion, it may not be the result of skillful activity. In fact, Hands et al. (2009) was able to show that while the low motor competence group was not less active, the group was significantly less aerobically fit. It is therefore possible that these children recorded a similar number of counts on the pedometer as the high competence group, yet at a reduced intensity.

It has been suggested that the hypoactivity that is often seen in children with DCD is linked to lower self-perception and poor self-adequacy (Hay, 1992; Hay & Missiuna, 1998). Likewise, it has also been observed that children with lower self-perceptions of their abilities in physical activity have poorer coordination and report lower levels of physical activity than their peers (Wrotniak et al., 2006). Perceptions of poor general physical ability not only negatively affect performance, but may also make the activity less enjoyable (Cairney et al., 2006a). This negative cycle increases the likelihood that the child with DCD will choose to spend less time engaged in physical activity, and more time in pursuits for which they feel a sense of competence and enjoyment (Poulsen et al., 2007a). Reduced levels of participation in physical activity may also result as a consequence of mechanically inefficient movement patterns. Children with DCD are likely to experience earlier fatigue than well coordinated individuals as a result of mechanical inefficiency (Hands & Larkin, 2002). Children with DCD have also been reported to have a less efficient running technique than their typically developing peers (Larking & Hoare, 1991), which may also accelerate fatigue and reduce time spent engaging in physically active pursuits and sports.

2.5. Limitations and Methodological Challenges

There are gaps in the literature evident from this review that require attention in future research on children and adolescents with DCD. First, large scale epidemiologic longitudinal studies that quantify risk over time and changes in health outcomes are lacking. Only eight studies have utilized longitudinal follow-up designs. However, short-term follow-up durations in some of these studies make it impossible to know how the impact of DCD changes from childhood to adolescence and in particular, what the consequences of poor motor competency are on the health and well-being of children as they progress into adulthood. Measuring change over time also presents some difficulty, as standardized tests for assessing DCD such as the BOTMP and the MABC are primarily designed to measure motor proficiency in school-aged children. Currently, there is no single instrument that covers the entire range of motor problems that may be present in adolescents (Cantell et al., 2003). An extensive multi-level instrument is required to assess developmental change in the adolescent population. In studies on long-term outcomes, the validity of the case ascertainment and outcome measures are imperative with respect to the sensitivity to change in the variables under investigation.

In the absence of a ‘gold standard’ to identify the presence of DCD, the reviewed studies relied on different instruments and assessment protocols for classifying subjects. Only two studies in this review applied the full DSM-IV diagnostic criteria to classify children with DCD. While some authors acknowledge the feasibility of using the DSM-IV criteria, in practice, its use is poor, and attempts have been made to address this problem (Sugden, 2006). Consistent definitions are still needed for samples to be comparable across studies. Furthermore, heterogeneity in the case ascertainment

instruments, and differences in the cut-offs used for the same test instruments hindered direct comparisons between the various samples in the reviewed studies. Crawford et al. (2001) investigated the performance of children with varying motor skills on the BOTMP, MABC and DCDQ, and found that it was not unusual for subjects to score within the average range on one test while classified as a case on another. In fact, the overall agreement between tests was reported to be less than 80%. Moreover, the percentile used as a cut-off point heavily influences the severity of the cases included in the study sample. The cut-off should always be reported and taken into account when interpreting results. Another issue to consider is the type of subjects that are included in the comparison group. Not having a control or comparison group, as children “with” the condition and those without are typically only separated by a cut-off set by the investigator, is a common problem in DCD research. Clearly, when children in the lowest motor proficiency centile are compared against those in the highest centile, differences between groups are exaggerated, as opposed to when all children, with varying motor abilities (low, moderate, high) are used in the analysis.

Additional methodological challenges are associated with assessment of children’s fitness and physical activity. These constructs are generally difficult to operationalize and measure in a population with typical motor skills. Children with DCD may have added challenges such as performing the test properly and may also be more vulnerable to psychological impacts of the assessment itself. For example, field tests of aerobic fitness such as the shuttle run are typically conducted in a group setting, where children with motor difficulties may find these assessments particularly stressful due to low perceived adequacy toward physical activity (Cairney et al., 2006a). Other studies

have shown that children with DCD have greater anxiety associated with participation in motor tasks (Rose et al., 1994; Schoemaker and Kalverboer, 1994), which may also negatively impact on their test results. The same concern can be found in lab-based testing (Silman et al., in press).

Our systematic review was limited to English language literature; potentially missing other relevant international publications. Also, we chose not to formally appraise the quality of the evidence, as we considered all research designs and study types as significant contributions to this review. Ideally, studies with greater methodological rigour (e.g., experimental versus observational designs) should be given greater weight when synthesizing evidence. As such, our review aimed to capture a broad spectrum of study types with heterogeneous methodologies and varying methodological strengths, and did not attempt to quantify or pool estimates across studies.

2.6. Summary

The results of this systematic review demonstrate that motor competence plays an important part in fitness and physical activity outcomes. It has been clearly demonstrated that body composition, cardiorespiratory fitness, muscle strength and endurance, anaerobic capacity, power, and physical activity have all been negatively associated, to various degrees, with poor motor proficiency. However, differences in flexibility were not conclusive as the results on this parameter are mixed. Overwhelmingly, cardiorespiratory fitness, muscle strength and endurance, and physical activity outcomes are negatively affected by poor motor skills. For most fitness components, performance levels were significantly lower in the DCD population. It is well understood that fitness

is related to health itself, and low fitness levels may compromise health and well-being. These results also highlight the concern that children with DCD are at risk for poor cardiovascular health, which is developed and maintained through regular participation in physical activity (Faught et al., 2005). In fact, independent contributions of fitness and physical activity towards risk for health and cardiovascular disease have been established (Strong et al., 2005). Moreover, poor aerobic fitness early in life may have important consequences for the development of cardiovascular disease later on (Berenson, 2002). Likewise, regular physical activity is supported in clinical and epidemiologic research to minimize the risk of chronic disease and to maximize well-being, with the benefits clearly demonstrated in pediatric populations (Sothorn et al., 1999).

There are methodological challenges associated with the assessment of children's fitness and physical activity, and various measurement tools are often used to assess the same construct (e.g., aerobic fitness), making comparisons across studies difficult. It is also debatable whether it is more important to measure children's participation in physical activity or their level of physical fitness (Angilley & Haggas, 2009). The impact of physical activity on fitness in any cohort is rate limited as much as the fitness level is predetermined through genetic predisposition (Bouchard et al., 1997). However, physical activity participation is a behaviour that is influenced by a multitude of factors, including the decision to incorporate an active lifestyle, self-perception, social pressures, and environmental and physical constraints among others (Cermak & Larkin, 2002). In children with poor motor skills, the ability to be active is compromised while decreasing the opportunity to develop health-related fitness. It is important to gain a better understanding of the factors that influence children's participation and how patterns of

physical activity and physical fitness are created to provide information critical for the design of appropriate activity-based interventions. Future work should also consider other aspects of physical fitness over time, especially since outcomes such as muscle endurance in addition to aerobic fitness will have important implications regarding the capacity to be physically active.

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CHAPTER 3 – A Prospective Cohort Study Comparing Workload in Children with and without Developmental Coordination Disorder²

3.1. Introduction

Developmental coordination disorder (DCD) is a prevalent childhood condition characterized by motor coordination difficulties that affect day-to-day activities such as dressing, feeding, and writing (Wilson, 2005). DCD is thought to affect approximately 5-9% of school-age children (APA 2000; Gillberg & Kadesjo, 2003; Cermak & Larkin, 2002). The cause of DCD has not been established; however, it is generally believed to be a chronic impairment that persists into adulthood (Barnhart et al., 2003; Cantell et al., 1994).

Children with DCD are at risk for overweight/obesity, lower overall fitness levels, poor perceived physical competence, lower activity levels, and reduced motivation to participate in physical activity (Cantell, Smyth, & Ahonen, 2003; Cairney et al., 2005; Cairney et al., 2007; Poulsen, Zivinai, & Cuskelly, 2008; Schott et al., 2007). In light of the increasing prevalence of cardiovascular disease risk factors observed in children and adolescents, those for whom compromised motor proficiency presents challenges for engaging in physical activity may be of particular concern. One of the many consequences of reduced physical activity is that health-related fitness components such as cardiorespiratory fitness (CRF) are compromised (Hands, 2008). Higher levels of CRF have been associated with numerous health benefits, whereas poor fitness is an independent risk factor for a variety of negative health outcomes, including

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cardiovascular disease, and premature mortality (Katzmarzyk et al., 2004). While previous work has shed light on these disconcerting patterns for children with poor motor proficiency, many gaps in the literature exist and large scale, prospective, longitudinal, studies that quantify disease risk in this population of children are still lacking (Rivlis et al., 2011a).

A widely used direct assessment of cardiorespiratory fitness is performed by measuring peak oxygen uptake (peak VO_2) during a maximal exercise test. This requires measurement of respiratory gas exchange by indirect calorimetry and is performed in a controlled laboratory environment. Field tests such as the 20-m shuttle run, that measure CRF responses, are frequently used as a proxy, particularly in large community-based samples where individual laboratory assessments are not feasible. A recent systematic review reported that children with DCD had, on average, 11-22% lower $\text{VO}_{2\text{peak}}$ using lab-based assessments, and 17-28% lower aerobic capacity in field-based tests (Rivlis et al., 2011a).

Very few prospective studies have been conducted that describe the long-term trajectories of cardiorespiratory fitness in children with DCD relative to children without motoric difficulties. In particular, differences in CRF as children progress into adolescence are not well understood. Considering the importance of CRF as a key determinant of future health status, we sought to assess how CRF changes over time, and to delineate factors that may have an impact on CRF in children with DCD. In order to isolate the independent effect of DCD, we consider gender, BMI, school, perceptions of self-efficacy (adequacy), and physical activity participation as covariates.

In a previous publication, we compared CRF differences between children with DCD and their peers in a 2.5 years prospective follow-up study (Cairney et al., 2010a). In the current investigation, we add to previous findings by following the same cohort of children into adolescence, for a total surveillance period of approximately five years. Given the longer follow-up period in the current study and the increased number of observations, we now have the ability to see if the observed trend continues, to control for confounding factors (e.g. perceptions of adequacy), and to examine three way interactions (e.g., between DCD, gender, and time). The outcome we are using in the current study is maximum speed attained during the final stage of the 20-m shuttle run. The measure has not been transformed in any way, and therefore is less prone to bias that may be associated with using a formula to calculate peak VO_2 in children (Fairbrother, Jones, & Hitchen, 2005; Penry, Wilcox, & Yun, 2011; Ruiz et al., 2009; Stickland et al., 2003). Using the non-transformed shuttle run results also allows us to estimate the relative impact of factors such as BMI on overall test performance on the shuttle run. This is not possible when using the allometrically scaled transformation, which scales the test results to body composition (weight in kg).

3.2. Methods

3.2.1. Data collection

This study is part of a prospective cohort follow up designed by the Physical Health Activity Study Team (PHAST). The PHAST is a longitudinal investigation following a large cohort of children from Grade Four to Nine in the District School Board of Niagara (DSBN). The project began in September 2004 with all students enrolled in

Grade Four (average age=9.9 yrs at baseline). A total of 2278 children from an original sample of 2378 (representing 75 of 92 possible schools) agreed to participate in annual school-based health assessments (95.4% consent rate). In the autumn of 2004, the pilot phase of PHAST took place, where we established testing and training protocols, developed a cadre of trained assistants, and completed baseline testing. The first formal wave of data collection took place in the spring of 2005. Subsequent assessments were conducted bi-annually (i.e., autumn and spring of each school year) for 2005 through 2007. In 2008-2009, due to the addition of a laboratory-based component to the study (reported elsewhere) and pedagogic concerns of the school board, only one annual school-based assessment was possible. Overall, eight waves of data collection were carried out (not including the pilot phase in year 1) over the course of 56 months. The number of subjects available for analysis for each wave of data collection, as well as subjects' characteristics are reported in Table 3-1. Research ethics approval was provided by Brock University and the DSBN.

3.2.2. Motor proficiency and case ascertainment

Children's motor proficiency was evaluated using the short form of the Bruininks–Oseretsky Test of Motor Proficiency (BOTMP-SF), using standardized procedures (Bruininks & Bruininks, 2005). The short form has been previously validated for school-age children against the full-scale test with high correlations (Bruininks, 1978). The short form contains 14 items that examine general motor skills including running speed and agility, balance, bilateral coordination, strength, upper-limb coordination and dexterity, and response speed. Sampling procedures are reported in

detail elsewhere (Cairney et al., 2010a). In brief, motor assessments were conducted by a team of trained research assistants in each school's gymnasium. Children who scored at or below the tenth percentile (based on population derived norms) on the BOTMP-SF were classified as probable DCD (pDCD) for all analyses. This cutoff score has been used in previous research to define probable cases (Cairney et al., 2007) and corresponds to population-based estimates of the prevalence of the disorder (between 5% and 9%) (Gillberg & Kadesjo, 2003; Hay, Hawes, & Faight, 2004). We describe cases as probable DCD because our primary means of case identification is through results of a field test administered by researchers, not a full diagnostic protocol administered by a physician. Moreover, our method does not include all criteria stipulated in the Diagnostic and Statistical Manual of Mental Disorders, version IV (American Psychiatric Association, 2000). Criterion B (limitations in activities of daily living) was not determined which is not atypical of research in this area as most studies do not take into account the exclusion criteria in the DSM-IV (Visser, 2003). Although the BOTMP-SF does not provide an in-depth analysis of each aspect of motor proficiency, it does provide an excellent assessment of general motor functioning.

3.2.3. Cardiorespiratory fitness

Aerobic capacity was assessed using the Léger 20-m shuttle run test. This test is a validated, well-established field measure of maximal oxygen uptake in children (Léger & Gadoury, 1989). Students are required to run back and forth on a 20m track at a progressively increasing pace (0.5 km/h every minute), controlled by signals from a standardized compact disk recording. These assessments were conducted in the school

gymnasium, where subjects performed the assessment in groups of approximately 10-15 students. The test was terminated when a child could not maintain the required running pace for two consecutive 20m segments. The maximal speed attained during the final stage of the test was subsequently used as the dependent measure in the analysis (Flouris et al., 2005; Léger & Gadoury, 1989).

In this study, we chose not to use the predicted VO_2 max equated from the Léger shuttle run as a measure of test performance, selecting stage completed instead. While the 20 m shuttle run test is an acceptable field assessment tool for cardiorespiratory fitness, its original prediction equation may be prone bias. Newer prediction models that increase the accuracy in evaluating VO_2 peak have been proposed (Flouris et al., 2005). It has also been suggested that gender distinct equations provide more accurate prediction of VO_2 max from 20-m shuttle run test (Stickland et al., 2003). Furthermore, studies have shown the Léger equation to underestimate VO_2 max values (Fairbrother, Jones, & Hitchen, 2005; Penry, Wilcox, & Yun, 2011; Ruiz et al., 2009). Stage completed is a measure that is independent of weight, and is closer to an estimation of workload. Using workload as opposed to VO_2 reduces concerns over the accuracy of equations used for predicting VO_2 max. Furthermore, the statistical analysis strategy used in this study allowed us to control for any potential confounding due to weight and/or gender, among other covariates.

3.2.4. Physical activity and perceptions of adequacy

The Children's Self-Perceptions of Adequacy in, and Predilection for Physical Activity (CSAPPA) scale was administered to measure self-perceptions of generalized self-efficacy toward participation in physical activity. Items are structured in an

alternative choice format presenting gender and culture neutral descriptions of different aspects of physical activity (Hay, 1992). The CSAPPA contains sub-scales for adequacy, predilection, and enjoyment of physical education class. The Participation Questionnaire (PQ) was also administered to provide a measure of children's physical activity participation. The PQ contains 63 items that provide a frequency estimate of children's participation in the areas of free-time play and recreation, intra-mural sports, inter-school sports, community sports teams and clubs (i.e., swimming, tennis), and sport and dance lessons (Hay, 1992). Participation in organized activities covers a 1-year period, and free-play is recalled from typical pastime choices. Higher scores indicate a greater number of active choices or "activity units". The PQ has consistently demonstrated strong construct validity and test-retest reliability (Hay 1992; Hay, Hawes, & Faught, 2004).

3.2.5. Statistical analysis

To explain CRF changes over time in children with and without pDCD, a mixed effects model was fitted. The model takes into account the nesting of observations within children over time (i.e., repeated measures) and the nesting of children within schools. Random intercepts at the school and student levels were estimated. Since we modelled a growth curve with unequally spaced data, the spatial power SP(POW) covariance structure was specified (Littell et al., 2006). We examined the contribution of different covariates of interest on the change in variance in the dependent variable. In order to test whether trajectories of CRF differed between children with pDCD and typically developing children over the course of the investigation, the model examined the main effects of pDCD, time, and the interaction between the two, adjusting for relevant

covariates (e.g., gender, BMI, activity level, predilection for activity). We also tested for a three-way interaction between pDCD, time, and gender given that boys have, on average, higher CRF than girls, and that previous work has shown boys with pDCD to be at greater risk of poor CRF than both typically developing children and girls with pDCD (Wu et al. 2010). All analyses were conducted using SAS version 9.1

Table 3-1. DCD status, sex, and Léger running speed by wave

	Grade 4	Grade 5	Grade 6	Grade 7	Grade 8	Grade 9		
	Wave 1	Wave 2	Wave 3	Wave 4	Wave 5	Wave 6	Wave 7	Wave 8
Participants (n)	2278	2229	2229	2117	2125	1807	1707	1581
BOTMP-SF Tested	2002	2046	2046	2018	2020	1440	1369	1221
Probable DCD	7.89%	7.58%	7.58%	7.58%	7.52%	7.08%	7.30%	7.04%
% Male (n)	50.9% (1155)	50.8% (1128)	50.8% (1128)	50.4% (1066)	50.5% (1073)	50.6% (915)	50.4% (860)	51.7% (817)
Max Léger Running Speed (km/h)								
No DCD								
Mean (SD)								
Male	10.25 (0.87)	10.49 (0.94)	10.64 (0.94)	10.61 (1.00)	10.67 (1.10)	10.93 (1.16)	11.27 (1.19)	11.47 (1.13)
Female	9.84 (0.69)	10.04 (0.76)	10.21 (0.78)	10.20 (0.80)	10.25 (0.89)	10.47 (0.95)	10.52 (0.93)	10.45 (0.91)
pDCD								
Mean (SD)								
Male	9.25 (0.42)	9.41 (0.61)	9.49 (0.75)	9.46 (0.68)	9.51 (0.77)	9.51 (0.81)	9.66 (0.75)	10.13 (1.01)
Female	9.27 (0.43)	9.31 (0.41)	9.37 (0.47)	9.39 (0.52)	9.39 (0.44)	9.60 (0.55)	9.69 (0.65)	9.63 (0.66)

3.3. Results

3.3.1. Univariate statistics

Baseline sample characteristics and descriptive statistics are provided in Table 3-1. Trajectories of the outcome measure maximum Léger running speed are depicted in Figure 3-1. There were no group differences in height over time, suggesting that the rate of growth in the pDCD and the control group was similar and any changes in the outcome measure are not likely to be due to maturational differences. Our results showed that while both groups demonstrated an increase in running speed over time, children with pDCD had consistently lower values relative to controls, with pDCD females demonstrating the lowest scores over time. Also, we observed that the magnitude of the difference in run speed increased over time. The mean group difference between boys with pDCD and those without increased from 1.00 at baseline to 1.34 at the final assessment point (a difference of 34%), while for girls, the difference increased from 0.57 at baseline to 0.82 at the final follow up (a difference of 44%). Repeated measures analysis of variance indicated an overall gender and pDCD status difference, with run speed scores for girls significantly lower than those for boys ($F=273.21$, $p<0.0001$) and for children with probable DCD compared to those without the disorder ($F= 242.92$, $p<0.0001$).

3.3.2. Multivariate model

The results of the multivariate, mixed effects analysis are presented in Table 3-2. We found a significant main effect of pDCD ($\beta=-0.8254$, $p<0.0001$) when controlling for gender, BMI, activity level, and predilection for activity. The negative and significant

estimate for pDCD indicates that children with pDCD had a lower maximum run speed at any given wave, when controlling for relevant covariates. We also tested whether the relationship between run speed and pDCD changed over the course of the investigation, since the interaction term between pDCD status and wave was significant (fixed effect $F=-2.96$, $p=0.0042$), suggesting that there was a difference in trajectories of CRF between children with and without the disorder. Furthermore, we found evidence that the trajectories of run speed in children with probable DCD and those without differed by gender. In other words, we found a significant three-way interaction between probable DCD status, wave and gender. As depicted in Figure 3-1, overall CRF levels were lower in girls than boys at any given time, and both males and females in the control group consistently outperformed those with pDCD.

Figure 3-1. Maximal Léger run speed attained over time for children with pDCD and controls

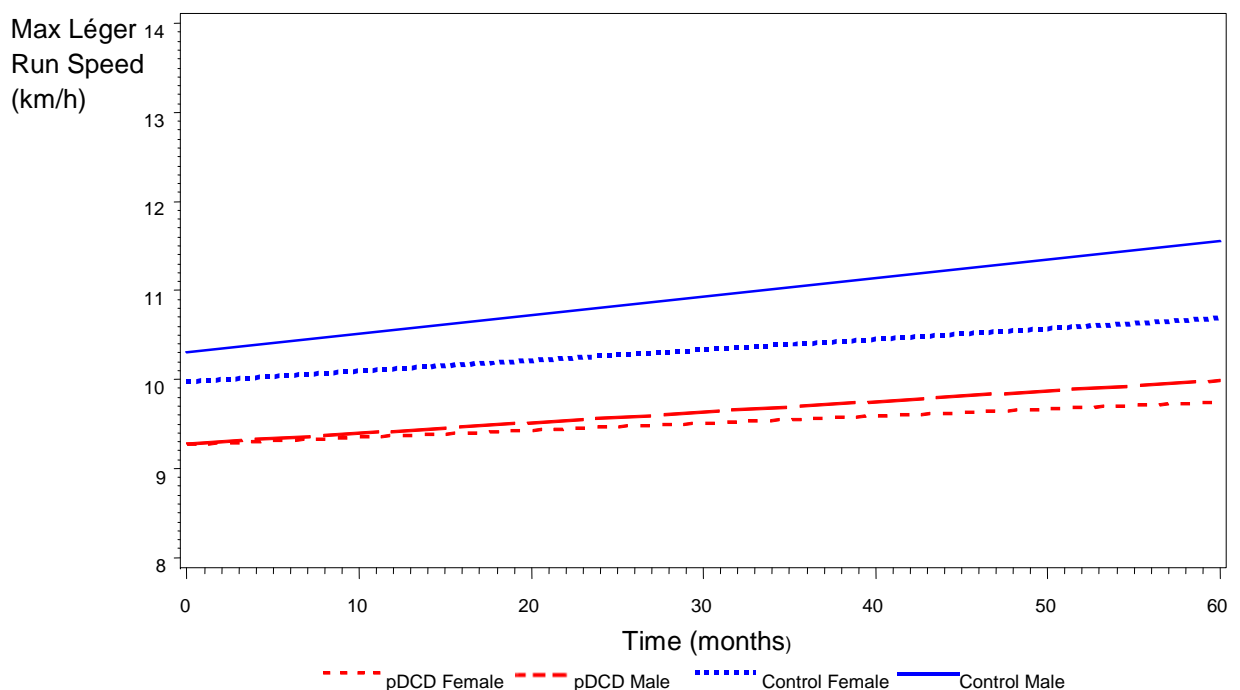


Table 3-2 Mixed Effects Model Results Predicting Maximal Léger Run Speed

	Estimate	SE	t	p Value
Intercept	10.6952	0.1253	85.38	<.0001
Female	-0.2814	0.03082	-9.13	<.0001
BMI	0.000787	0.01448	-28.33	<.0001
PQ	0.008948	0.001077	8.31	<.0001
CSAPPA	0.01448	0.000787	18.40	<.0001
pDCD	-0.4529	0.05508	-8.22	<.0001
Wave	0.02599	0.000632	41.10	<.0001
Wave*Female	-0.00822	0.000727	-11.31	<.0001
pDCD*Wave	-0.00716	0.001673	-4.28	<.0001
pDCD*Wave*Gender	0.006371	0.002102	3.03	0.0024

3.4. Discussion

The purpose of the present study was to examine trajectories of CRF, measured here as maximum Léger run speed, in children with and without probable DCD. Although previous work has found differences in CRF between these groups (Cairney et al., 2007; Castelli & Valley, 2007; Haga 2008), data have been cross-sectional, with CRF assessed at a single point in time. Few studies have reported CRF changes over time in these populations (Cairney et al. 2010a, Haga, 2008; Hands, 2008). These studies were based on either small samples comparing children with low and high motor competence, e.g. n=67 in Haga (2008), and n=38 in Hands (2008), or did not have comparably long follow-up duration (e.g., 2.5 years Cairney et al., 2010a). Ours is the first study in the literature to examine workload differences related to probable DCD longitudinally over a

relatively long surveillance period (five years) among a large cohort of children. Furthermore, we were able to follow children as they entered adolescence, strengthening our understanding of the relationship between CRF and DCD in this older cohort of children.

Our results are consistent with previous studies, in that we found substantial differences in CRF between children with and without pDCD at baseline. Moreover, these group differences not only persisted over the study period, but we also observed a small yet significant increase in the decline in run speed between groups over time. In other words, children with pDCD had a slower rate of increase in CRF with age compared to typically developing peers. We observed no difference in the rate of growth (in height) between the pDCD group and the controls, therefore it is unlikely that maturational differences or body size could explain these results. Similar to previous work, we found evidence of a gender interaction between pDCD and CRF over time. Girls with pDCD had the lowest levels of CRF throughout the study, however both genders with probable DCD showed a greater rate of decline in CRF over time. Boys with pDCD seemed to perform consistently worse not only relative to typically developing boys, but also compared to girls without the disorder.

With regard to children without motoric difficulties, our Léger run speed results are remarkably similar to previous research on children in this age range. Olds (2006) presented a meta-analysis of 109 studies that have used the 20-m shuttle run to assess global, age- and sex-specific means in children from 37 countries. In typically developing 10 year olds, the average final speed was reported to be 10.46 km/h for boys and 9.96 km/h for girls, while in our study the final speed was 10.49 and 10.04 km/h, respectively.

Similarly, at 14 years old, Olds (2006) reports an average run speed for boys of 11.52 km/h, while in our study the boys averaged a final speed 11.47 km/h. Although the group of 14 year old girls in our study seemed to slightly outperform the average, with the final run speed of 10.45 km/h vs. the global average of 10.31 km/h. It should be noted that Canadian children as a group were found to perform slightly better than the overall average (SD=0.245 deviation from the global age- and sex-specific mean).

A few contributing factors that may be potential explanations for the difference in CRF observed between children with and without pDCD are overweight and lack of physical activity. It has been shown that poor motor proficiency is clearly associated with higher BMI and lower levels of physical activity and participation in free and organized play (Rivlis et al., 2011a). However, in our study we were able to account for these factors by controlling for differences in BMI and physical activity in our analysis. Another factor to consider as potentially influencing CRF differences is the oxygen cost of locomotion and mechanical efficiency. Children with poor motor proficiency have been reported to have a less efficient running technique than their typically developing peers (Larking & Hoare, 1991). Rivlis et al. (2011b) reported that while exercising to exhaustion on a cycle ergometer, children with pDCD required greater relative oxygen uptake at any given submaximal workload relative to children without pDCD. It is possible that poor technique while performing an aerobic fitness test is responsible for the increased oxygen cost and an earlier onset of fatigue in children with poor motor proficiency. This may explain why these children are unlikely to persist at a running task and may give up sooner on tests of endurance.

As with any study, there are limitations that need to be addressed when evaluating the results. One concern relates to the type of methodology used to assess CRF. A potential limitation with field-based measures of aerobic capacity such as the shuttle run, is that it relies on the internal motivation of the child to perform to exhaustion. For children with DCD this is particularly challenging, as they generally perceive themselves to be less competent and may have less motivation to continue the assessment, potentially dropping out prematurely, underestimating the true aerobic capacity of this group (Cairney et al., 2006; Silman et al., 2011). However, in the laboratory setting, other indicators such as heart rate (HR) and respiratory exchange ratio (RER) can be used to monitor a child's performance to ensure a true maximum is achieved (Silman et al., 2011). Cairney et al. (2006) suggest that at least part of the reason children perform less well on tests of aerobic endurance is because they do not believe themselves to be as adequate as other children at physically active pursuits. In fact, they found that one-third of the effect of DCD on shuttle run performance can be attributed to differences in perceived adequacy. Taking this into account, we were able to address this difference in our study by controlling for the effects of perceived adequacy in the multivariate model.

A closely related concern is whether children with DCD can adequately perform the shuttle run test, given their coordination difficulties. Although the shuttle run test was designed to require minimal motor competence, it does demand pacing and rapid turning, which could pose challenges for some children with DCD. At the same time, laboratory-based protocols, which require the child to cycle or run on a treadmill, could also pose challenges. Cairney et al. (2010b) addressed the issue of comparability of the shuttle run and the lab based cycle ergometer tests for assessing CRF, showing moderate to good

correlations between the two tests. Nonetheless, the findings of the laboratory based studies corroborate those reported in field tests, suggesting a true deficit exists and that the effect of DCD on aerobic capacity is not limited by the type of methodology used to assess CRF. However, choosing a test suitable to the motoric capabilities of subjects with DCD may be a useful protocol to adopt in future studies although likely difficult to implement in practice since motoric challenges in DCD vary widely from child to child. Another limitation is that Figure 3-1 represents raw data and does not reflect trajectories estimated from the mixed model, therefore the results may look somewhat different if plotted using the predicted parameter estimates from the model.

Our results show that the difference in maximum Léger run speed between children with and without probable DCD is substantial, and that it tends to increase over time. The trends observed here suggest that children with pDCD are more likely to fall into the low fitness range at a much faster rate than typically developing children. It is well understood that fitness is related to health itself, and low CRF levels may compromise both health and well-being. These results also highlight the concern that children with DCD are at risk for poorer cardiovascular health, which is developed and maintained through regular participation in physical activity and aerobic activities (Faught et al., 2005). This adds to the argument suggesting that interventions intended to improve physical fitness may be appropriate and necessary for children with motor difficulties. This is particularly important as the hypoactivity and poor perceived physical competence cycle, as described in children with DCD by Bouffard et al. (1996), tends to be established in childhood, and often persists into adulthood. It is recommended that future work address the factors that influence aerobic fitness and how patterns of physical

activity and physical fitness are created in children with poor motor competence to provide information critical for the design of effective interventions.

3.5. References

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CHAPTER 4 - Oxygen Cost During Incremental Exercise Test in Children with Developmental Coordination Disorder and Controls³

4.1. Introduction

Developmental coordination disorder (DCD) is a prevalent childhood condition characterized by motor coordination difficulties that affect a child's ability to perform day-to-day activities such as dressing, feeding, and writing (Larkin & Hoare, 1991). Engaging in physical activity presents a further challenge for children with DCD. This is supported by evidence that children with DCD are less likely to participate in organized sport and free play (Rivlis, Hay, Cairney, Klentrou, Liu, & Faught, 2011). It has been consistently demonstrated in previous work that children with DCD are disadvantaged to various degrees on most fitness parameters including: body composition, aerobic power, muscle strength, endurance, anaerobic power, and physical activity (Hands, 2008; Hands & Larkin, 2002). A particular component of fitness strongly correlated with health is aerobic power, and this is consistently lower in children with DCD (Cairney, Hay, Veldhuizen, & Faught, 2009; Wu, Lin, Li, Tsai, & Cairney, 2010). A systematic review of literature reported that children with DCD had on average 11-22% lower VO_2peak using lab based assessments, and 17-28% lower aerobic power in field based tests such as the 20m shuttle run test (Rivlis, Hay, Cairney, Klentrou, Liu, & Faught, 2011). Furthermore, the decline in aerobic power in children with DCD is more rapid over time compared to typically developing children (Rivlis et al., 2011).

³ A version of this chapter has been submitted for publication in *Developmental Medicine & Child Neurology*

While some studies have compared VO_2peak in children with DCD to that of typically developing children, there are no reports of the submaximal oxygen cost during an incremental test in this cohort. Baerg, Cairney, Hay, Rempel, Mahlberg, & Faught (2011) reported significantly higher energy expenditure in children with DCD during habitual physical activity as measured by a seven day accelerometry assessment, suggesting the possibility that energy demands are greater in children with motoric challenges during varying intensity levels of daily physical activity. However, a recent study, which examined the oxygen cost of walking and running at standardized speeds of children with and without DCD, failed to demonstrate any differences - possibly due to small effect sizes and limited sample size (Chia, Guelfi, & Licari, 2009). In a recent study Silman, Cairney, Hay, Klentrou, & Faught (2011) suggested that the effect on VO_2peak could be accounted for by the negative consequences of DCD, such as poor movement patterns resulting in higher energy expenditure and higher levels of fatigue. Although they were unable to test the submaximal differences, the authors speculated that even slightly compromised movement efficiency in children with p-DCD may have contributed to increased energy demands in early stages of the VO_2peak test. We sought to extend this work in the current study.

Examining submaximal oxygen cost (submaximal VO_2) can contribute to our understanding of the differences in aerobic power between children with and without DCD. This is important when considering that most activities of daily living are performed at submaximal endurance levels. Therefore, the purpose of this study is twofold: 1) to compare the submaximal oxygen cost of children with and without DCD during an incremental exercise test on a cycle ergometer and 2) to identify how

submaximal oxygen intake differs with increasing workloads between children with and without DCD.

4.2. Methods

4.2.1. Research design and participants

This nested case-control design was an ancillary study as part of a larger longitudinal investigation by the Physical Health Activity Study Team (PHAST) (Cairney, Hay, Veldhuizen, Missiuna, & Fought, 2010a). From the initiation of the study in 2004, we measured the motor coordination of 2260 children in annual, school-based assessments using the short form Bruininks–Oseretsky test of Motor Proficiency (BOTMP-SF) (Crawford, Wilson, & Dewey, 2001). To recruit participants for the lab-based component of the current study, telephone calls were initiated in the Fall of 2007 to 115 of the 2260 children (5.1%) who were identified at risk of DCD (BOTMP-SF rank at or below 10th percentile), as well as 115 control subjects matched on age within three months, gender and school. The study was approved by the Research Ethics Boards for both Brock University and the District School Board of Niagara. All subjects and their care-givers provided informed written assent and consent, respectively.

4.2.2. Assessment of motor proficiency

A total of 147 subjects accepted an invitation to participate in a laboratory component of the PHAST longitudinal cohort investigation. These subjects included 67 suspected cases of DCD based on cut-off of >10% on their motor coordination scores using the BOTMP-SF and 80 controls. All subjects were re-assessed using the Movement Assessment Battery for Children, 2nd edition (MABC-2), which measures

both gross and fine motor coordination (Henderson & Sugden, 2007) and the Kaufman Brief Intelligence Test, 2nd edition by a trained occupational therapist blind to the child's BOTMP-SF score. The MABC-2 is the most frequently used standardized motor test to screen for children with DCD and is recognized as being both reliable and valid (Crawford, Wilson, & Dewey 2001; Tan, Parker, & Larkin, 2001). Consistency between BOTMP-SF and MABC-2 was found in 79% (53/67) of cases and 86% (69/80) of controls. A priori power calculations suggested a minimum of 60 subjects in each group. Therefore, subjects who screened into the laboratory as controls, but who scored below the 16th percentile on the MABC-2 were classified as cases for the laboratory investigation. Matches based on age, gender and school location were possible for 63 cases of probable DCD and 63 controls (26 female pairs and 37 male pairs).

Criterion A of the Diagnostic and Statistical Manual of Mental Disorders states that motor coordination should be substantially below that expected for the person's age and intelligence (APA, 2000). It should be noted that a full assessment of all criteria to confirm a diagnosis of DCD was not possible; specifically, that impairment significantly interferes with academic achievement or activities of daily living. We have chosen to use the term probable DCD (pDCD) to acknowledge this limitation.

4.2.3. Measures

Relative body fat was assessed using whole body air-displacement plethysmography (BOD POD Life Measurement, Inc, Concord, CA) (Fields, Hunter, & Goran, 2000). The BOD POD has repeatedly demonstrated to be a reliable and valid technique in evaluating body composition in children and obese individuals (Nunez,

Kovera, & Pietrobelli, 1999). Prior to the assessment, subjects were instructed to avoid any movement, to relax and breathe normally while inside the chamber. The surface of clothing and hair has a significant impact on volume measurements, therefore, all subjects wore tight fitting swimsuits or spandex shorts and a swim cap. Body volume was measured twice, each session lasting approximately 40 seconds, with the two results averaged. Subject pubertal maturity was self-assessed using the appropriate Tanner staging pictures (Taylor et al., 2001). Physical activity was measured with an Actical accelerometer, which has been established as a valid measure step-count, activity counts (Puyau, Adolph, Vohra, Zakeri, & Butte, 2004) and activity energy expenditure (Everson, Catellier, Gill, Ondrak, & McMurray, 2008) in healthy children. Children were fitted with the accelerometer (Actical, version 2.0, Mini Mitter, Respironics, 2006) to wear for a 7-day period following their visit to the laboratory where aerobic assessments were conducted. Parents were provided with a log to record the time within each day that their child had removed/replaced the unit for bathing, swimming, and bedtime. Activity was recorded in 30-second epochs for total daily activity counts for each day. The Children's Self-Perceptions of Adequacy in, and Predilection for Physical Activity (CSAPPA) scale, as well as the Participation Questionnaire (PQ), which contains 63 items that provide a frequency estimate of children's physical activity, were also administered (Hay, 1992).

4.2.4. Assessment of oxygen cost

Using a continuous, incremental exercise protocol, submaximal and peak aerobic power ($\text{VO}_{2\text{peak}}$) were measured on a cycle ergometer (Excalibur Sport V2, Lode BV, Groningen, Netherlands). Each subject had a practice period of a few minutes on the cycle ergometer to familiarize themselves with the equipment. The saddle, handle and

pedals of the cycle ergometer were adjusted to give optimal comfort and efficiency for the subject while peddling. Initial power output was set at 20-40 Watts (depending on gender, weight) for the first three minutes warm-up period, after which work rate was increased by 20 Watts every minute until the final stages. During the estimated final stages, 15 Watt increments were used until volitional fatigue. Subjects were instructed to keep a constant cycling speed of 60 to 65 rpm for the duration of the test. Throughout the assessment, all subjects were verbally encouraged. This protocol is similar to that used in a recent study to assess oxygen cost of children during submaximal exercise on a treadmill (Reybrouck, Vangesselen, Mertens, & Gewillig, 2007). As soon as the test finished, subjects gave a rating of perceived exertion (RPE) using a standardized Borg scale (Borg, 1998). Heart rate was recorded continuously during the test and metabolic gases were analyzed using an AEI metabolic cart (Model S-3A, AIE Technologies, Pittsburgh, Pennsylvania). The criteria used to verify the achievement of peak aerobic power were two of the following: 1) respiratory gas exchange ratio of at least 1.00, 2) heart rate >85% of age-predicted maximal heart rate, or 3) signs of intense effort, e.g. hyperpnoea, facial flushing, or difficulties maintaining the required speed of the cycle (Armstrong & Van Mechelen, 2008). For comparison of differences between the groups, the measured, absolute values of $\text{VO}_{2\text{peak}}$ were then normalized for body size (body mass in kilograms) and fat free mass (FFM in kilograms).

4.2.5. Statistical analysis

All statistical analyses were performed using SAS (version 9.2). First, descriptive statistics were calculated including mean and standard deviation for subject's age, relative body fat, BMI, activity count and peak exercise performance data. In order to

assess submaximal performance, analysis was carried out on data collected between 25%-75% of maximum intensity, which represented a range of workloads of 25 to 165 Watts. To test whether trajectories of VO_2 differ between children with and without pDCD over the course of the incremental exercise protocol, a mixed effects or hierarchical model (Brown & Prescott, 2006) was used to examine the main effects of pDCD, workload, and the interaction between the two, on the dependent variable - oxygen cost (VO_2), as measured during submaximal exercise in ml/kg/min. Mass-related VO_2 is widely used to express oxygen uptake since VO_2 is strongly correlated with body mass. This is conventionally controlled for by dividing VO_2 (ml/min) by body mass (kg) and expressing it as the simple ratio ml/kg/min (Armstrong, Tomkinson, & Ekelund, 2011). The model was also adjusted for relevant covariates including gender, relative body fat, BMI, physical activity participation, CSAPPA, and activity count. To control for the subjects' heterogeneous maximal aerobic power profiles, we included $\text{VO}_{2\text{peak}}$ as a predictor in the model. To estimate change in VO_2 over time with incremental increases in workload, adjusting for correlation of measures within subject; the mixed model procedure REPEATED statement was used. Matched case-control pairs were entered as a random effect in the model (Brown & Prescott, 2006). Since the different levels of the repeated effect represent change over time; a time series component within each subject (a first order autoregression) was incorporated into the model. Assumptions of the model were verified to ensure that the chosen analysis strategy was appropriate for the data.

4.3. Results

Participants' characteristics are provided in Table 4-1. Children with pDCD had significantly higher Body Mass Index (BMI) (23.4 ± 5.9 vs. 20.2 ± 4.0 , $p < 0.0006$) and

relative body fat (28.3% vs. 20.0%, $p<0.0001$) compared to children without the disorder. Children with pDCD were also significantly less active than children without the condition, with an average activity count of 176,865 (± 57535) compared to 210,949 (± 76068) in the control group ($p<0.0084$). Pubertal maturity did not differ between the two groups.

VO₂peak was significantly lower in the pDCD group, even when maximal aerobic power was normalized to fat free mass (Table 4-2). There were no significant differences in maximum heart rate between groups. Furthermore, children with pDCD had similar rates of perceived exertion values as the controls. The average respiratory exchange ratio (RER) exceeded 1.0 in both groups, although it was significantly higher in control subjects ($p<0.0045$).

Table 4-1. Physical characteristics of study participants (mean \pm SD)

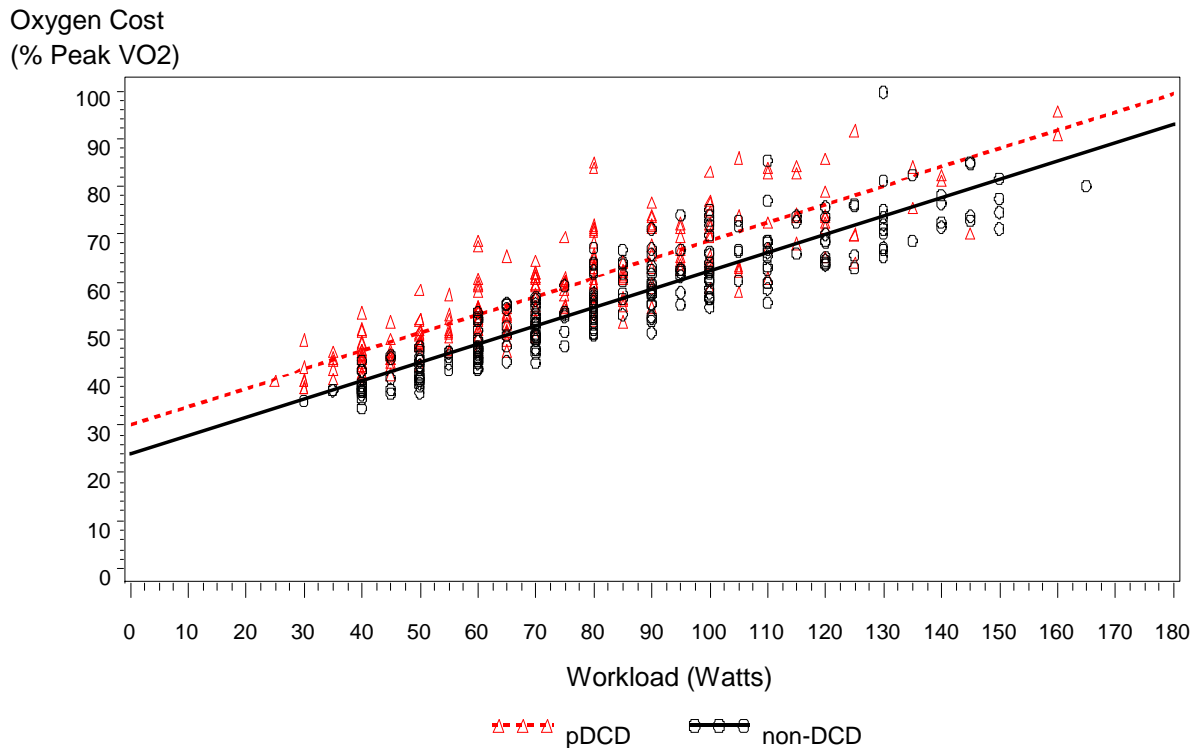
Characteristic	pDCD (n=63)	non-DCD (n=63)	P-value
Male	37	37	NA
Female	26	26	NA
Age (years)	12.9 (0.44)	12.8 (0.38)	NA
Body Mass Index (kg/m ²)	23.4 (5.9)	20.2 (4.0)	0.0006
Relative Body Fat (%)	28.3 (11.2)	20.0 (9.87)	<.0001
Activity count (counts/day)	176,865 (57535)	210,949 (76068)	0.0090

Table 4-2. Peak exercise performance data (mean \pm SD)

Characteristic	pDCD (n=63)	non-DCD (n=63)	P-value
VO ₂ peak (ml/ffm in kg/min)	48.8 (7.3)	53.2 (8.1)	0.0017
VO ₂ peak (ml/kg/min)	35.0 (7.66)	42.9 (8.06)	<.0001
Final RER	1.05 (0.09)	1.10 (0.09)	0.0045
Maximum HR (beats/min)	187.5 (14.6)	191.6 (12.5)	0.0960
Final RPE	16.9 (2.01)	17.0 (1.52)	0.9177

The relative oxygen cost expressed as a percent of $\text{VO}_{2\text{peak}}$ is shown in Figure 4-1. Controlling for individual variation in maximal oxygen uptake by plotting values as a percentage of $\text{VO}_{2\text{peak}}$ demonstrated that children with pDCD had consistently greater oxygen cost at any given exercise intensity (Figure 4-1). The regression equation for cases was: $\text{VO}_2 = 35.47 + 0.300 \cdot \text{Watts}$. While for the controls the regression equation was: $\text{VO}_2 = 30.27 + 0.297 \cdot \text{Watts}$. In this plot, the intercept was significantly different ($p=0.0364$), but the slopes did not differ ($p=0.918$).

Figure 4-1. Predicted relative oxygen cost (expressed as a percent of peak VO_2) as a function of workload



The results of the multivariate, mixed effects analysis presented in Table 4-3. The dependent variable in the model, (VO_2 ml/kg/min), showed a significant main effect for pDCD ($\beta=3.1888$, $p=0.0006$), when controlling for relative body fat, $\text{VO}_{2\text{peak}}$ and workload. While other variables were considered in the modeling process (e.g., gender, a self-reported measure of participation in active play, generalized self-efficacy toward physical activity, and activity count), these were not significant and therefore not included in the final model. The positive and significant estimate for pDCD indicates that cases had a higher oxygen cost at any given submaximal workload, when controlling for relevant covariates. We also tested whether the relationship between workload and pDCD changed over the course of the incremental exercise test. We found the interaction term between pDCD status and workload to be significant ($p=0.0004$).

Table 4-3. Results of mixed effects model for the outcome submaximal oxygen cost (ml/kg/min)

Variable	Estimate	SE	T-value	P-value
Intercept	-0.4938	2.7608	-0.18	0.8586
Relative body fat (%)	-0.1055	0.03698	-2.85	0.0046
$\text{VO}_{2\text{peak}}$	0.2494	0.04899	5.09	<.0001
Workload (Watts)	0.1939	0.006054	32.04	<.0001
pDCD	3.1888	0.9224	3.46	0.0006
Workload*pDCD	-0.03151	0.008820	-3.57	0.0004

4.4. Discussion

This study demonstrated a significant relationship between pDCD and oxygen cost at submaximal workloads in children. The results of this study confirm that differences in levels of motor coordination in children affect oxygen cost. Children with pDCD require greater relative oxygen uptake at any given submaximal workload. It was observed that, over the course of the incremental exercise protocol, children with pDCD were consistently working at a greater percentage of their VO_2peak . This indicates that they were exercising at a higher metabolic rate to sustain the same level of workload relative to children without pDCD.

To our knowledge, this is the first study to assess the oxygen cost of cycling at submaximal workloads in children with DCD. Previous research has compared the oxygen cost of locomotion using treadmill walking and running in children with DCD (Chia, Guelfi, & Licari, 2009), and in other populations with greater motor difficulties, such as children with cerebral palsy (CP) (Maltais, Pierrynowski, Galea, & Bar-Or, 2005). However this study was the first to demonstrate that children with pDCD have greater oxygen costs at submaximal exercise intensities. Even at very low workloads (less than 40 Watts), at the very beginning stages of the exercise protocol, we found children with pDCD to be disadvantaged as they demonstrated a higher relative VO_2 compared to controls. It is possible that even at very low intensities children with poor motor proficiency need to utilize more energy to carry out the basic movements associated with maintaining proper posture and position on the cycle ergometer.

Energy expenditure and oxygen uptake are influenced by an individual's fitness level. Corroborating earlier studies, we found children with pDCD to have a lower

VO₂peak than controls (Cairney, Hay, Faught, Flouris, & Klentrou, 2007; Cairney, Hay, Veldhuizen, & Faught, 2010b). Variation in maximal oxygen uptake was controlled for in the analysis by entering VO₂peak for each subject as a covariate in the mixed effects model. As expected, it was found that body fat percentage was a significant predictor of oxygen cost, which was adjusted in the multivariate model to tease out the effect of pDCD. Physical activity (PQ and activity count) and CSAPPA variables were not significant predictors of VO₂ in the model. Although there is some evidence to show that perceived adequacy towards physical activity may contribute to the difference in VO₂ between pDCD and typically developing children (Silman, Cairney, Hay, Klentrou, & Faught, 2011). However, the literature is contradictory as to whether physical activity and VO₂peak in children are related (Armstrong, Tomkinson, & Ekelund, 2011).

Furthermore, in order to test whether trajectories of VO₂ differed between children with and without pDCD over the course of the exercise protocol, we examined the interaction between pDCD and workload. A significant interaction between pDCD and workload was observed, which indicated that the difference in VO₂ between cases and controls at higher workloads is greater than that at lower workloads ($p=0.0004$).

The oxygen cost of work performed is frequently used in the literature as a proxy measure of movement efficiency (Karlsen, Helgerud, Stoylen, Lauritsen, & Hoff, 2009; VanSwearingen et al., 2009). While we did not set out to specifically measure movement efficiency in the current study, our findings shed some light on this issue. It is hypothesized that an individual who consumes more oxygen to sustain a set level of activity has a higher energy cost relative to someone who consumes less oxygen for the same level of activity, and is therefore less efficient and more likely to perform poorly in

aerobic activities (Chia, Guelfi, & Licari, 2009; Maltais, Pierrynowski, Galea, & Bar-Or, 2005). While very few studies have attempted to explore the subject of oxygen cost and its determinants in children with DCD, literature on children with other motoric disorders, such as those with Cerebral Palsy (CP) can help elucidate this relationship. For example, children with CP have been shown to have a greater oxygen cost of walking compared to typically developing children, with higher oxygen costs associated with increased severity of CP (Maltais, Pierrynowski, Galea, & Bar-Or, 2005; Johnston, Moore, Quinn, & Smith, 2004). Corroborating our results, previous studies reported that children with CP tend to walk at a higher relative exercise intensity, or percentage $\text{VO}_{2\text{peak}}$, compared with the more 'economical' form of typically developing individuals (Dahlback & Norlin, 1985; Maltais, Pierrynowski, Galea, & Bar-Or, 2005).

While Baerg et al. (2011) did not examine oxygen cost, but energy expenditure, it was suggested that energy demands during habitual physical activity are greater in children with poor motor proficiency given their significantly higher expenditure. In another study that examined the oxygen cost of locomotion using treadmill walking and running (Chia, Guelfi, & Licari, 2009), it was noted that children with DCD appeared to be working at a higher relative percentage of their $\text{VO}_{2\text{peak}}$ at all four speeds used in the assessment compared to those without DCD. The study, however, failed to demonstrate a significant difference in the oxygen cost of walking or running between those with and without DCD. Chia et al. (2009) hypothesized that this may have been due to differences in movement proficiency between children with and without DCD being insufficiently large to affect oxygen cost, and potentially, a small sample size ($n=31$). The authors also noted that subjects who held onto the railing for support during running were excluded

from the analysis, which represented a large proportion of the DCD group. Therefore, the study effectively included those who were more capable of walking and running on a treadmill, which may have minimized any potential differences between groups and limited the generalizability of the findings. Another limitation of the study was that not all the children met the criteria for achieving $\text{VO}_{2\text{peak}}$. In contrast, in the current study, we are confident that a true maximum effort was obtained from the participants as evidenced by the RER, max HR values, and RPE ratings. This is important for calculations of relative VO_2 , specifically where a percent of $\text{VO}_{2\text{peak}}$ was used (e.g. Figure 4-2). Moreover, we found that even when the energy to support weight is not required, as is the case with the cycle ergometer (in contrast to the treadmill protocol), we still found significant differences in oxygen cost between children with and without DCD.

Factors that are purported to affect oxygen cost of movement in individuals with motor difficulties have been proposed to include poor muscle strength, low muscle tone, muscle co-contraction, spasticity, and inefficient energy transfers between body segments (Johnston, Moore, Quinn, & Smith, 2004; Unnithan, Dowling, Frost, & Bar-Or, 1996). However, sparse empirical evidence exists, and few studies have addressed this issue in children with DCD. There is also a possibility that the internal cost of exercise may be higher in children with DCD, this is supported by the higher intercept for the children with pDCD in Figure 4-2 in the current study. Another factor to consider as potentially influencing energy expenditure is joint mobility. According to Hands (2008), children with poor motor proficiency tend to have heterogeneous fitness profiles, which may result in extreme ranges of flexibility or rigidity. Kirby and Davies (2007) showed

similarity in symptoms seen in some DCD children to those with joint hypermobility syndrome. Children with poor motor proficiency have also been reported to have a less efficient running technique than their typically developing peers (Larkin & Hoare, 1991). It is possible that poor technique while performing the test is responsible for the increased oxygen cost. However, this is challenging to measure in practice, and greater attention should be paid in the future to elucidate this relationship more clearly.

A possible implication of consistently working at a greater percent of $\text{VO}_{2\text{peak}}$, while performing the same activity as children with typical motor proficiency, is that the child with DCD is more likely to have an accelerated onset of fatigue. Exercise can be perceived as more strenuous. In fact, children with DCD are likely to experience earlier fatigue than well coordinated individuals as a result of inefficient or wasteful movements (Hands & Larkin, 2002; Reybrouck, Vangesselen, Mertens, & Gewillig, 2007). This is a troubling consequence as it may manifest in reduced time spent engaging in physically active pursuits and sports, as children with DCD may find it challenging to keep up with motorically proficient peers. Physical activity entails the additional expenditure of energy, which, for children with coordination difficulties, may exacerbate the already elevated demands placed on the body even at low intensity aerobic activities.

4.4.1. Limitations

A limitation that needs to be addressed in future work relates to the methods used in the current study. Although the incremental cycle ergometer test is a valid assessment of submaximal and $\text{VO}_{2\text{peak}}$ in children (Armstrong, 1998; Reybrouck, Vangesselen, Mertens, & Gewillig, 2007), comparing submaximal oxygen cost between groups can be

performed under greater control at steady states. We were not able to achieve this due to the study design. Therefore, it is important to verify the findings of this study using more stringent laboratory assessments.

4.5 Conclusions

It was demonstrated that differences between levels of motor coordination in children affect oxygen cost. Children with pDCD utilized more oxygen to sustain the same level of submaximal workload relative to typically developing controls and therefore perform closer to their maximal exercise ability at each level of workload. This may be an important factor to consider as children typically spend the majority of their time performing at submaximal intensities. A possible implication of this is that the child with DCD is more likely to have an earlier onset of fatigue compared to those without the disorder. In particular, the disadvantage of poor motor proficiency in children with DCD should be explored in future studies as a potential factor contributing to greater oxygen cost of movement.

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CHAPTER 5 – General Discussion

5.1. Overview

This thesis explored several questions related to the broad topic of physical activity and fitness in children with DCD. First, a systematic review examining the literature on fitness and physical activity patterns in children with DCD was performed to provide an update of the recent literature and to synthesize relevant data. The next chapter examined the longitudinal trajectories of cardiorespiratory fitness (CRF) of children with and without pDCD in a prospective cohort design. The last chapter reports a study that compared the oxygen cost of exercise at submaximal workloads between children with pDCD and matched, typically developing controls. The thesis utilized the PHAST dataset, which has been prospectively following up children from the District School Board of Niagara since 2004 to examine their fitness and physical activity patterns, motor coordination deficits, and corresponding risks for cardiovascular disease. The research presented in this thesis is the culmination of this six year longitudinal examination. A total of 2278 children enrolled in Grade 4 at baseline (representing 75 of 92 possible schools) agreed to participate in the PHAST annual school-based health assessments. From this larger sample, a subset of 126 subjects participated in the lab-based component of the study. In this chapter, the main research findings of the thesis are summarized and the implications of these findings discussed. The chapter concludes with suggestions for future research.

5.2. Overall Thesis Findings

Results of this thesis provide an important contribution to our understanding of DCD. In Chapter 2, several fitness parameters were reviewed using evidence from recent literature, and the differences between typically developing children and those with DCD were quantified. Aerobic fitness was consistently deficient in children with DCD. Children with DCD had, on average, 11-22% lower VO_2peak using lab-based assessments, and 17-28% lower CRF in field-based tests. This is particularly concerning as poor aerobic fitness is linked to poor health (Twisk, 2002). Research has also demonstrated that higher levels of aerobic fitness are associated with a healthier cardiorespiratory profile in children and adolescents (Ortega, 2008). In addition, poor aerobic fitness early in life may result in the development of cardiovascular diseases later in life (Berenson et al., 2002, Twisk 2002). Other findings highlighted in Chapter 2, were the higher levels of body fat and the consistent hypoactivity of children with pDCD relative to their typically developing peers.

Based on the first study of this thesis, and the negative consequences of poor aerobic fitness, it was recommended that future studies focus on the longitudinal trajectory of CRF over time in children with pDCD. Subsequent to the recommendations of the systematic review, in Chapter 3 of this thesis, we report a study of a large group of children followed over time in order to understand how CRF changes relative to typically developing children. This study showed that group differences in aerobic fitness not only persisted over the study period of almost five years, with the pDCD group showing a consistent deficit, but a small yet significant increase in the decline in CRF between

groups over time. Furthermore, changes in aerobic fitness over time may be different depending on gender, where girls with the disorder showed the largest deficits.

As discussed previously, studies examining factors associated with aerobic performance in children with and without DCD are lacking. Research is needed to enhance our understanding of the reasons for aerobic deficits in children with DCD. To this end, Chapter 4 presents the first evidence to indicate potential sources of bioenergetic differences underlying DCD. Although the results of this thesis cannot provide direct support for reduced energetic efficiency as a determinant of poor aerobic performance in this population of children, the findings suggest that this hypothesis warrants further investigation. Biomechanical assessments and exercise testing under steady state conditions may be helpful in future studies designed to test the degree of energetic inefficiency associated with DCD compared to typically developing children. Furthermore, this thesis sets the groundwork for further work that can contribute to our understanding of the factors that affect exercise and aerobic performance in children with DCD, which may be useful in designing appropriate interventions.

5.3. Deficits in Fitness and Physical Activity in Children with DCD

No comprehensive systematic review has been carried out to date on the fitness and physical activity patterns of children with DCD. The novel contribution of this work is that it revealed clear trends in outcomes, as such information is not easily gleaned from individual studies which often report widely varying results. Two clear findings emerged from the systematic review presented in Chapter 2. First, children with poor motor proficiency generally have poorer performance than their peers on most measures of

physical fitness, including: body composition, CRF, muscle strength and endurance, anaerobic capacity, and power. However, differences in flexibility were not conclusive as the results on this parameter are mixed. Second, these children are generally less physically active than their peers. The review summarized the deficits of the various fitness parameters available from the literature by providing quantitative ranges for each parameter where available, and comparing the findings of relevant studies.

For body composition, the evidence overwhelmingly supported an increased risk for elevated body fat in children with DCD, with the majority of studies (13 out of 18) showing significant differences between groups. Moreover, the magnitude of the difference in BMI between DCD and typically developing children ranged as high as 40% (e.g. Silman et al. 2011). A few of the studies that did not find significant differences examined younger cohorts of children. For example, a sample of 5 to 7 year old children in the study by Hands (2008) did not demonstrate differences in BMI. Similarly, Schott et al. (2007) and Williams et al. (2008) did not find associations between DCD and BMI in 4–9 year old and 3–4 year old children, respectively. It is arguable that the detrimental effect of poor coordination on body composition does not manifest itself until late childhood to early adolescence as increases in body fat are cumulative in nature.

This is a noteworthy finding, as in early childhood children can become increasingly more involved in physical activities and organized sports. Those with poor motor coordination are potentially at greater risk for inactivity and as a result are more prone to weight gain and obesity. Furthermore, an increase in BMI and adiposity may directly affect children's performance on activities such as running, and jumping,

independent of the effect of DCD, due to the energetic disadvantage of excess weight and a higher oxygen cost of locomotion (Hands & Larkin, 2002, chap. 11). This emphasizes the importance of engaging children with DCD in physical activity early on, before the risk of excess adiposity creates a more challenging situation.

Another explanation for the lack of a significant association between body composition and DCD in younger cohorts could be due to the measurements used in the studies. BMI was the most common measure used and it appears to have poor sensitivity in screening for overweight children (Mast et al., 2002). Therefore, the relationship between body composition and motor competence could be obscured. It is recommended that in future studies, body composition analysis (e.g., fat mass and distribution) be considered for screening children at risk of becoming obese as BMI may not be a sensitive enough measure.

The review also pointed out that a clear deficit in CRF is associated with poor motor proficiency. In fact, 18 of the 19 studies reported that children with DCD had decreased aerobic capacity compared to their typically developing peers. Children with DCD had on average 11–22% lower VO_{2peak} using lab-based assessments, and 17–28% lower CRF in field-based tests (e.g., 20 meter shuttle run). Furthermore, evidence from the few longitudinal studies on CRF reviewed in Chapter 2 suggests that the negative effects of poor motor proficiency persist as children mature in age (Cairney et al., 2010a; Haga, 2009; Hands, 2008). However, the finding that CRF declined at a greater rate in the DCD group (i.e., an interaction effect with time) was not consistently observed. For example, Haga (2009) found a main effect of DCD, however no interaction effect with time was evident, potentially due to either the small sample size ($n = 67$) or the short

follow-up (32 months). This highlights the need for larger studies with more frequent test points and longer follow-up durations to better appreciate the changes in CRF as children progress into adolescence. To this end, Chapter 3 of this thesis provides evidence that, in a large cohort of children, CRF is not only consistently lower in those with DCD, but that this deficit between the two groups increases over time.

In addition to the deficits in CRF experienced by children with DCD, the review identified 14 studies that examined muscle strength and endurance, all of which reported a negative effect of low motor proficiency on this parameter. This provides convincing evidence that a child with DCD is likely to suffer from the consequences of poor muscle strength to some degree. Adequate muscle strength and endurance are important for performing many daily activities and sports without fatigue. Poor muscle strength may result in poor posture, musculoskeletal problems such as lower back pain or lax joints, and difficulty participating in sports, particularly those requiring force production (Hands & Larkin, 2002). Practitioners working with children diagnosed with DCD should explore the use of strength training as part of an intervention strategy to improve the adverse outcomes associated with poor muscle strength (Kaufman & Schilling, 2007).

The review in Chapter 2 also examined the effect of poor motor proficiency on levels of physical activity and participation in free and organized play. Poor motor proficiency was adversely associated with these outcomes in 20 of the 21 reviewed studies. As discussed in Chapter 2, there are challenges when comparing physical activity across studies due to the various measures used to assess the construct of physical activity. In most studies, self-report questionnaires were used. The use of more objective measures, such as accelerometers or pedometers was uncommon and these studies

reported smaller effect sizes. The use of accelerometry to measure physical activity offers improvements over self-report techniques. However, it should be noted that there is currently no clear consensus on the scoring and interpretation of accelerometry data in measuring physical activity behaviour (Ward et al., 2005). The review also considered trajectories of physical activity, although only four longitudinal studies assessed participation over time. Taken together, the results of the longitudinal studies suggest that children with DCD are less likely to participate in free play or organized activities consistently over time. However, unlike CRF the physical activity deficit does not appear to increase with age.

5.4. Consistent Cardiorespiratory Deficit in Children with DCD Persists into

Adolescence

Chapter 3 of this thesis is the first study to examine workload differences related to DCD longitudinally over a relatively long surveillance period (4.7 years) among a very large cohort of children. In this study, we were able to follow children as they entered adolescence, strengthening our understanding of the relationship between CRF and DCD in this cohort of children. Few studies have reported CRF changes over time in this population. Although previous work has found differences in CRF between DCD and non-DCD groups (Cairney et al., 2007; Castelli & Valley, 2007; Haga 2008), most data have been cross-sectional, with CRF assessed at a single point in time. In addition, previous studies were based on either small samples comparing children with low and high motor competence, (Haga, 2008; Hands, 2008), or shorter follow-up durations (e.g., 2.5 years in Cairney et al., 2010a).

Our five year longitudinal cohort study showed that while both groups demonstrated an increase in running speed over time, children with pDCD had consistently lower values relative to controls. Also, it was observed that the magnitude of the difference in running speed increased over time. The mean group difference between boys with pDCD and those without increased from baseline to the final assessment point by 34%, while for girls, the difference increased by 44%. These results suggest that children with pDCD tend to fall behind the typically developing group in their CRF, and that the difference is even more pronounced in girls with pDCD.

We also tested for a three-way interaction between pDCD, time, and gender given that boys have, on average, higher CRF than girls, and that previous work has shown boys with pDCD to be at greater risk of poor CRF than both typically developing children and girls with pDCD (Wu, Lin, Li, Tsai, & Cairney, 2010). We found a significant three-way interaction between probable DCD status, time, and gender, in other words, the trajectories of aerobic performance in children with pDCD and those without differed by gender over time. However, it seemed that girls with the disorder were more disadvantaged in CRF over time compared to boys in this study.

In Chapter 4 we reported that while exercising to exhaustion on a cycle ergometer, children with pDCD required greater relative oxygen uptake at any given submaximal workload relative to children without pDCD. Considering the differences in shuttle run performance, it is possible that poor technique while performing an aerobic fitness test, such as the shuttle run, is responsible for the increased oxygen cost and an earlier onset of fatigue in children with poor motor proficiency. This may explain why these children are unlikely to persist at a running task and may give up sooner in

endurance activities. Furthermore, mixed effects modeling demonstrated that the effect of pDCD on shuttle run performance was significant even when BMI was taken into account. This finding suggests that the effect of motor incoordination is not due solely to children with DCD being heavier than their typically developing peers, but that the difference can be attributed to other factors associated with DCD.

5.5. Children with DCD Require More Oxygen to Perform the Same Workload

Relative to Peers

To our knowledge, Chapter 4 is the first study to assess the oxygen cost of cycling at submaximal workloads in children with pDCD. Previous research compared the oxygen cost of locomotion using treadmill walking and running in children with DCD (Chia, Guelfi, & Licari, 2009), and among other populations with greater motor difficulties, such as children with cerebral palsy (CP) (Maltais, Pierrynowski, Galea, & Bar-Or, 2005). However this study was the first to demonstrate that children with pDCD have greater oxygen cost while cycling at submaximal exercise intensities.

As discussed in Chapter 1, several authors have hypothesized that inefficient movement may account for the challenges experienced by children with DCD when engaging in physical activity. Chapter 4 provided physiological evidence that this may indeed be the case, as these children consistently consume more oxygen at submaximal workloads relative to typically developing peers. While the study in Chapter 4 was not designed to test the effect of mechanical inefficiency on aerobic performance, findings from this study support the hypothesis that children with DCD are disadvantaged in aerobic activities due to their greater oxygen cost of movement, which is likely caused by

biomechanical inefficiencies. It has been speculated that ‘wasteful’ movements, such as those associated with poor motor proficiency, for example, muscle co-contraction, low muscle tone, hyperflexibility, and spasticity, contribute to greater energy expenditure during aerobic activity (Johnston, Moore, Quinn, & Smith, 2004; Unnithan, Dowling, Frost, & Bar-Or, 1996). It was shown in Chapter 4 that in this group of children greater energy expenditure, as determined by oxygen cost, was needed to sustain the same workload on the cycle ergometer relative to controls. This lends support to the hypothesis that the incoordination experienced by children with DCD may play a role in hampering movement economy.

It has been suggested in a study of children with mild CP that the poor movement economy in this group of children might be one cause of their early fatigability (Maltais et al., 2005). Those with low cycling economy would be working at a higher relative exercise intensity or percentage peak VO_2 than more economical individuals, and have less “metabolic reserve,” and earlier fatigue. Considering the greater oxygen cost of movement in this population and the prevalent hypoactivity (as shown in Chapter 2), low physical activity levels associated with poor motor proficiency may be a compensatory mechanism to reduce or prevent fatigue. This finding is supported in a study by Maltais et al. (2005) that reported a strong negative linear relationship between physical activity levels and the oxygen cost of walking in children and adolescents with mild cerebral palsy. While the deficits in children with CP are different from those in children with DCD, the underlying mechanism that may impact fatigue could be comparable (e.g. higher oxygen cost of movement). It should also be noted this deficiency in children with DCD in our study was observed while they performed a very structured cycle ergometry

test as opposed to less structured movement activities such as running. Therefore, economy of movement may be even more compromised during regular daily physical activity motor requirements.

Teachers and peers often perceive children with DCD as less physically active even though their physiological load, as measured by oxygen consumed, may actually be higher (Cermak & Larkin 2002). Since the movement patterns of these children are poorly coordinated and inefficient, higher energy demands may be required to perform routine tasks that their healthy peers take for granted (Cermak & Larkin 2002). This thesis suggests that children with DCD may also be at risk of experiencing early fatigue even when executing aerobic exercise at very low level intensities. This increases the likelihood that the child with DCD will choose to spend less time engaged in physical activity, and more time in pursuits for which they feel a sense of enjoyment which will not require a high level of physical effort and subsequent fatigue (Poulsen et al., 2007a).

In Chapter 2, one of the suggestions for future research was to gain a better understanding of the factors that influence children's participation in physical activity, and to investigate how patterns of physical activity and physical fitness are created. The implication of these findings is that it provides practitioners with information critical for the design of appropriate activity-based interventions. This thesis provides evidence that the relative oxygen cost may be greater for children with DCD, which may contribute to earlier and greater fatigue and a decreased desire to engage in physical activity. These findings may be relevant to those working with children with DCD in helping to better understand the challenges associated with exercise in this population of children. In turn, interventions that help children become more energy efficient may make being physically

active more enjoyable. Children with DCD may require frequent rest periods when engaging in any degree of prolonged physical activity in order to avoid fatigue. Other factors to consider may be the use of muscle strengthening exercises, as this has been shown to lead to an increase in muscle tone, suggesting that energy costs could be lessened (Kaufman & Schilling, 2007). Although only a limited number of studies have addressed this issue, a study on the effect of physical training on the aerobic energy expenditure in physically handicapped children demonstrated that, after training, these children could perform the same intensity of exercise with lower energy expenditure (Dresen 1985). The potential for improving the economy of movement is important in these children because it can reduce symptoms of fatigue during exercise (Reybrouk, 2007).

5.6. Future Directions

Sufficient evidence exists to support the benefits of habitual physical activity in children (Sothorn et al., 1999; Twisk et al., 2002). Research presented in this thesis identified a consistent lack of physical activity in this population of children and quantified the degree to which children with DCD are less active than their peers. Furthermore, the finding that children with DCD tend to fall behind typically developing peers in aerobic fitness over time is a concern. Whether these patterns are carried over to adulthood remains unknown. It is well understood that increased physical activity is related to improved health outcomes, and that low physical activity and fitness levels compromise health and well-being (Strong et al., 2005). Since research suggests that

children with DCD are not likely to outgrow the condition (Cantell et al., 2003), these children appear at risk for persistent fitness deficits and hypoactivity as adults.

Our systematic review outlined several gaps in the literature that require further research among children and adolescents with DCD. First, it was noted that large scale epidemiologic longitudinal studies that quantify risk over time and changes in health outcomes are absent. This makes it impossible to know if and how the impact of DCD changes from childhood to adolescence and, in particular, the consequences of poor motor competency on the health and well-being of adolescents as they reach adulthood. Large scale, longitudinal studies spanning childhood, young adulthood, and beyond are essential if we are to fully understand the implications of this disorder. Future studies could probe the long term health impact resulting from physical inactivity, for example the risks of diabetes, cancer, and falling injuries in adults with DCD remains unknown.

It would also be pertinent to determine if there is any specific type of physical activity that children with DCD gravitate to. For example, swimming may be such an activity because it allows children to freely move without fear of falling, it is an individual sport, and also because it is a low-impact activity that generally has low injury rates associated with it. Parents and therapists working with children affected by DCD could encourage these types of activities to promote fitness and well-being.

While the research presented here did not set out to evaluate interventions for children with DCD, these findings provide useful information for the design of appropriate interventions for children with DCD and offer direction for future research. Specifically, we believe the increased oxygen cost of movement associated with DCD requires further investigation. The underlying mechanisms responsible for increased

oxygen cost of movement in children with DCD should be explored relative to biomechanical assessments. This has the potential to shed light on the factors that could be improved in those affected by poor motor competency. Future work could investigate strategies for improving the economy of movement in children with DCD during various types of daily activities. The next step would be to examine how these interventions relate to long-term functional and health outcomes.

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APPENDIX A – REB Letter of Approval

DATE: January 10, 2008

FROM: Michelle McGinn, Chair
Research Ethics Board (REB)

TO: Brent FAUGHT, CHSC
John Hay, John Cairney

FILE: 07-106 FAUGHT

TITLE: Establishing the Health Profile of Children with Motor Coordination Challenges

The Brock University Research Ethics Board has reviewed the above research proposal.

DECISION: Accepted as clarified

This project has received ethics clearance for the period of January 10, 2008 to December 30, 2011 subject to full REB ratification at the Research Ethics Board's next scheduled meeting. The clearance period may be extended upon request. *The study may now proceed.*

Please note that the Research Ethics Board (REB) requires that you adhere to the protocol as last reviewed and cleared by the REB. During the course of research no deviations from, or changes to, the protocol, recruitment, or consent form may be initiated without prior written clearance from the REB. The Board must provide clearance for any modifications before they can be implemented. If you wish to modify your research project, please refer to <http://www.brocku.ca/researchservices/forms> to complete the appropriate form Revision or Modification to an Ongoing Application.

Adverse or unexpected events must be reported to the REB as soon as possible with an indication of how these events affect, in the view of the Principal Investigator, the safety of the participants and the continuation of the protocol.

If research participants are in the care of a health facility, at a school, or other institution or community organization, it is the responsibility of the Principal Investigator to ensure that the ethical guidelines and clearance of those facilities or institutions are obtained and filed with the REB prior to the initiation of any research protocols.

The Tri-Council Policy Statement requires that ongoing research be monitored. A Final Report is required for all projects upon completion of the project. Researchers with projects lasting more than one year are required to submit a Continuing Review Report annually. The Office of Research Services will contact you when this form *Continuing Review/Final Report* is required.

Please quote your REB file number on all future correspondence.

MM/kw

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APPENDIX B – Child Letter of Informed Assent

Principal Investigators: Dr. John A. Hay, Brock University
Dr. John Cairney, University of Toronto and Brock University
Dr. Brent E. Faught, Brock University

Dear Parent and Child:

Thank you for your interest in our study. Please read the following information together. If you both feel comfortable and willing to participate in the tests described below, please check the boxes at the end of this consent form indicating child assent and parent consent.

Purpose: The purpose of this study is to look at healthy growth and development of children for the next three years.

Procedures: This assessment will take approximately 2.5 to 3 hours long and is divided into three parts. We thank you for participating. As promised, we have agreed to provide transportation for you to and from Brock University as well as \$50 for your family's participation in this study. Your participation is voluntary and you are free to withdraw from this study at any time without penalty from Brock University. Further, you are under no obligation to answer any or all questions or to participate in any aspect of this project. If you wish to stop participating in this study at any time, you and your parent will still receive free transportation from us as well as \$50 for your participation in the laboratory. Each part is described below.

PART I

This part of the study will be conducted in our laboratory at Brock University and requires 2.5 to 3 hours of your time. First, we would like you to complete the following forms, which will take about 10 minutes.

1. Medical Screening Questionnaire
2. Edinburgh Survey – Handedness Questionnaire

Next, we would like to complete a number of physical assessments on your child with the parent/guardian present. These assessments include:

1. **Body composition:**
 - a. Height and weight will be measured using a dual-purpose stadiometer.
 - b. 9 skinfold sites using painless pinch calipers. (It does not hurt).
 - c. Measure around the waist and hip using a flexible tape measure.
 - d. Bioelectric impedance analysis requires your child to stand on a weight scale and grasp handles. An electrical impulse travels from your child's hands to their feet. The impulse cannot be felt and causes no harm.
 - e. Lengths of your child's ring and index fingers.
 - f. Body muscle and fat weight will be measured while your child sits in the BOD POD chamber. If your child expressing previous or current anxiety for confined spaces, they will not be allowed to participate in this portion of the study. The BOD POD incorporates a built in window on the front of

the chamber in the event of a claustrophobic event or for communication purposes as well as a safety latch on the inside of the chamber for the subject to voluntarily exit on their own. During this 5-minute assessment, your child will be asked to relax and breathe normally.

2. **Cardiovascular health measures:** The carotid ultrasound method will be performed using a probe and pen like-devices. Heart rate will be measured using sensors placed on the skin of your child's chest. These sensors are used to detect the electrical activity generated by the heart and are not used to transmit electrical signals into their body from the heart rate monitor. Blood pressure is monitored using an automated arm cuff system that is similar to the method used in a doctor's office. A cuff is wrapped around the upper arm and is inflated then deflated. No risk is involved.
3. **Movement ABC² assessment:** This motor coordination assessment involving 8 short activities, including tasks such as tracing, cutting on a line and throwing a ball.
4. **Physical fitness assessment:** This assessment uses a bicycle to measure the maximum amount of heavy exercise. The bicycle tension will gradually get more difficult to pedal. A mask over the mouth and nose will be used to collect oxygen and carbon dioxide. The assessment will be finished when your child decides. One of the common risks of these kinds of assessments is the brief sensation of exhaustion. At the end of the assessment, your child will be asked to continue to pedal the bicycle at a very easy level until this sensation goes away. The risk of serious illness or death is extremely rare and is reduced by completing the medical screening questionnaire before the assessment and the continuous monitoring we will perform during the assessment.
5. **Accelerometer assessment:** This assessment will require your child to wear a small box the size of a smaller pager clipped onto their pant waist. The accelerometer is designed to measure activity movement that your child performs. We wish for your child to wear the accelerometer from the time they wake up, until they go to bed at night for 7 days. We also ask that the parent complete the Habitual Activity Estimation Scale and our Activity Log. There is no risk associated with this assessment. We will arrange to pick the accelerometer unit at your home.

PART II

The second part of the study would take place approximately 7 days from now at your home. We would come in the morning (before your child has breakfast) and it will only take about 10 minutes. We wish to collect a sample of your child's blood using a finger prick technique. The middle finger of your child's non-dominant hand (e.g. if they are right handed, we will use the middle finger of their left hand) will be pricked so two drops of blood can be sampled. Your child will feel a small prick, but will not feel any pain or discomfort for the remainder of the assessment. The tip of that finger may feel sensitive and a little bit sore for about a day. It is important to keep the site clean and covered with an adhesive bandage until it is healed to reduce the risk of infection. We will also use this moment to pick up the accelerometer that you will have had for the past week.

PART III

For this part of the study, we would like you to allow your child's homeroom teacher complete a survey on your child's combined listening, speaking, reading, writing, mathematics and reasoning skills. The name of this survey is the Learning Disabilities Diagnostic Inventory. Despite the name of this survey, we are not looking to diagnose any disabilities in your child's learning ability, nor are the teacher expected to provide a learning disabilities' diagnosis. We simply wish to see how able your child is while learning at school. The results of this assessment will not be shared with your child's school.

Participation and Withdrawal: Your child's participation is voluntary and they are free to withdraw from this study at any time without penalty from Brock University. Further, your child is not required to answer any or all questions or to participate in any aspect of this project.

Confidentiality: All personal data will be kept strictly confidential and all information will be coded so that your child is not associated with their answers. Only the researchers named above will have access to the complete data. Any information we receive will be entered immediately into computer records using a code number with no name attached. It is our intent to continue to publish the results of this research in scientific journals. Again, no personal information will be identified or be possible within any publication.

Information: This study has been reviewed and approved by the Brock University Research Ethics Board, (File#: 07-106) Research Services, Brock University, Room C315 - 905-688-5550 (Ext. 4315). We greatly appreciate your co-operation. If you would like to receive more information about the study, please contact **Dr. Brent E. Faught** at 905-688-5550, (Ext. 3586). If you are willing to grant permission to participate in this study, please complete the consent form below.

Thanks for your help!

Brent E. Faught, Ph.D.

John A. Hay, Ph.D.

John Cairney, Ph.D.

PARENT CONSENT FORM

I have read and understand the above explanation of the purpose and procedures of the project. My questions have been answered to my satisfaction.

- ☐ I give permission for my child to participate in **Part I** of the Brock University study conducted by Dr. John Hay, Dr. John Cairney and Dr. Brent E. Faught.
- ☐ As the participating child, I wish to participate in **Part I** of the Brock University study conducted by Dr. John Hay, Dr. John Cairney and Dr. Brent E. Faught.
- ☐ I give permission for my child to participate in **Part II** of the Brock University study

conducted by Dr. John Hay, Dr. John Cairney and Dr. Brent E. Faught.

- ☐ As the participating child, I wish to participate in **Part II** of the Brock University study conducted by Dr. John Hay, Dr. John Cairney and Dr. Brent E. Faught.
- ☐ I give permission for my child to participate in **Part III** of the Brock University study conducted by Dr. John Hay, Dr. John Cairney and Dr. Brent E. Faught.
- ☐ As the participating child, I wish to participate in **Part III** of the Brock University study conducted by Dr. John Hay, Dr. John Cairney and Dr. Brent E. Faught.

OR

- ☐ I do **NOT** give permission for my child to participate in the Brock University study conducted by Dr. John Hay, Dr. John Cairney and Dr. Brent E. Faught.
- ☐ As the participating child, I do **NOT** wish to participate in the Brock University study conducted by Dr. John Hay, Dr. John Cairney and Dr. Brent E. Faught.

Signature of Parent/Guardian: _____

Date: _____

Signature of Student: _____

Date: _____

APPENDIX C – Parent Letter of Informed Consent

Principal Investigators: Dr. John A. Hay, Brock University
Dr. John Cairney, University of Toronto and Brock University
Dr. Brent E. Faught, Brock University

Dear Parent/Guardian:

Purpose: The purpose of this study is to investigate healthy growth and development and its association with the physical activity of children for the next three years.

Procedures: We are requesting that you complete five forms as they relate to you and _____ (child's name). These forms will take approximately 40 minutes to complete.

Participation and Withdrawal: As a condition of your participation, we have agreed to provide transportation for you and your child to and from Brock University as well as \$50 for your family's participation in this study. Your participation is voluntary and you are free to withdraw from this study at any time without recourse from Brock University. Further, you are under no obligation to answer any or all questions or to participate in any aspect of this project. If you wish to discontinue participation in this study at any time, you and your child will still receive complementary transportation as well as \$50 for your participation in the study.

Confidentiality: All personal data will be kept strictly confidential and all information will be coded so that you are not associated with your answers. Only the researchers named above will have access to the complete data. Any information we receive will be entered immediately into computer records using a code number with no name attached. It is our intent to continue to publish the results of this research in scientific journals. Again, no personal information will be identified or be possible within any publication.

Information: This study has been reviewed and approved by the Brock University Research Ethics Board, (File#: 07-106) Research Services, Brock University, Room C315 - 905-688-5550 (Ext. 4315). We greatly appreciate your co-operation. If you would like to receive more information about the study, please contact Dr. Brent E. Faught at 905-688-5550, (Ext. 3586). If you are willing to grant permission to participate in this study, please complete the consent form below.

Thanks for your help!

Brent E. Faught, Ph.D.

John A. Hay, Ph.D.

John Cairney, Ph.D.

PARENT CONSENT FORM

I have read and understand the above explanation of the purpose and procedures of the project. My questions have been answered to my satisfaction.

- ☐ I wish to participate for the next three years in this Brock University study conducted by Dr. Brent E. Faught, Dr. John Hay and Dr. John Cairney.
- ☐ I do **NOT** wish to participate in this Brock University study conducted by Dr. Brent E. Faught, Dr. John Hay and Dr. John Cairney.

Signature of Parent/Guardian: _____

Date: